The impact of caregiving in Hoarding Disorder
Piloting a brief psychoeducational group for relatives of hoarders.

Thompson, Claire Amy Louise

Awarding institution:
King's College London

Systematic Review

Main Research Project

Service Related Project

Claire Thompson

Institute of Psychiatry, Psychology and Neuroscience

King's College London

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Institute of Psychiatry, Psychology and Neuroscience

King's College London

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Systematic review

A Systematic Review of Treatments for Hoarding Disorder

Supervised by:

Dr Juliana Onwumere

Dr Lorena Fernández de la Cruz

Professor David Mataix-Cols

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Abstract

Introduction: Hoarding disorder (HD) is a newly defined, OCD-related, mental health condition. Over the last decade, researchers have developed novel specialised treatments for problematic hoarding and, since the inclusion of HD in the nomenclature, of individuals fulfilling criteria for HD. The current study aimed to systematically review the treatments designed to improve HD symptoms and associated problems including anxiety, depression and functional impairment.

Method: An electronic search was conducted of PsycINFO, MEDLINE, EMBASE and Web of Science. Studies were included if: (i) the study evaluated an intervention for hoarding or an intervention for relatives of an individual with HD (ii) outcome measures were reported (including measures of hoarding symptoms or impact on life/distress levels, co-morbid psychiatric symptoms); and (iii) the paper was published in an indexed journal or published abstract from a professional/research conference. The quality of studies was assessed using the Clinical Trials Assessment Measure (Tarrier & Wykes, 2004). Results: 989 studies were identified through searches, from which seventeen studies met criteria and were included in the review, involving 474 participants with clinically significant hoarding symptoms or HD, and nine relatives. Treatments reviewed included cognitive-behavioural therapy, medication, cognitive remediation, and a relatives-only intervention. The majority of trials tested CBT in individual and group formats. Discussion: Most studies yielded statistically significant improvements in hoarding symptoms, although reductions were modest and many participants remained in the clinical range after treatment. Significant reductions were roughly equivalent after individual and group CBT, CBT combined with cognitive remediation, and a medication treatment. Quality assessment revealed that most studies were of poor quality and suggestions were made for future research which included: consistent measurement and diagnosis of HD, use of larger samples and randomised control designs with appropriate procedures to control for bias missing data, and inclusion of follow-up assessments.

1. Introduction

There has been an increasing amount of research into Hoarding Disorder (HD) in recent years, including the development of specialised treatments. However, questions remain about what treatments are most effective in treating the symptoms of HD, which can have a severe negative impact on individuals with HD and their families, including impaired individual quality of life, damaged family relationships and increased mortality risk (Frost, Steketee, & Williams, 2000; Saxena et al., 2011; Tolin, Frost, Steketee, & Fitch, 2008; Tolin, Frost, Steketee, Gray, & Fitch, 2008).

1.1 Defining Hoarding Disorder

HD has only recently been formally recognised as a mental health condition in its own right (DSM-5, American Psychiatric Association, 2013; see Appendix 1 for full diagnostic criteria) although research into the aetiology, characteristics, and treatment of hoarding problems dates back over two decades (Frost & Gross, 1993). HD was previously thought to be a relatively rare condition, but epidemiological research has now shown that clinically significant hoarding problems may affect as many as 2 to 5% of the adult population (Iervolino et al., 2009; Mueller, Mitchell, Crosby, Glaesmer, & de Zwaan, 2009; Samuels et al., 2008; Timpano, Exner, et al., 2011), and at least 1.5% would meet full DSM-5 criteria for HD (Nordsletten, Reichenberg, Hatch, Fernández De La Cruz, et al., 2013).

Formerly, hoarding was thought to be a symptom of obsessive-compulsive personality disorder (OCPD; American Psychiatric Association, 2000) or obsessive-compulsive disorder (OCD) (see Mataix-Cols & Fernández de la Cruz, 2014), and appeared as a compulsive symptom on one of the most widely used measures of OCD, the Yale-Brown Obsessive Compulsive Scale (Y-BOCS; Goodman, 1989). However researchers found that: (1) most people with hoarding symptoms did not meet criteria for OCD or OCPD (Frost, Steketee, & Tolin, 2011; Hall, Tolin, Frost, & Steketee, 2013; Mataix-Cols et al., 2013; Pertusa et al., 2008; Pertusa, Frost, & Mataix-Cols, 2010; Samuels et al., 2008); (2) hoarding could occur in other psychiatric conditions (Steketee & Frost, 2003); and (3) the presence of hoarding symptoms seemed to predict a poorer response to treatment for OCD compared to

people without hoarding (e.g., Bloch et al., 2014; Mataix-Cols, Marks, Greist, Kobak, & Baer, 2002; Mataix-Cols, Rauch, Manzo, Jenike, & Baer, 1999; Rufer, Fricke, Moritz, Kloss, & Hand, 2006). These findings led to HD eventually being defined as a distinct but OCD-related condition (Mataix-Cols et al., 2010; Pertusa, Frost, Fullana, et al., 2010). The DSM-5 diagnosis of HD excludes individuals with hoarding symptoms attributable to other disorders, such as Autism Spectrum Disorders or OCD (APA, 2013).

The primary symptom of HD is persistent difficulty discarding possessions (regardless of their actual value), due to a need to save items and distress experienced when attempting to discard items. The subsequent avoidance of discarding leads to an eventual build-up of clutter in the home, which at clinical levels compromises the ability for inhabitants to use rooms for their intended purpose (e.g., struggling to cook in the kitchen). For diagnosis, the hoarding would need to cause the person distress and/or impairment in social, occupational, or other aspects of functioning, which helps distinguish it from non-pathological collecting (Nordsletten, Fernández de la Cruz, Billotti, & Mataix-Cols, 2013). Over two thirds of people with HD also acquire possessions excessively through purchasing or collecting free items (Frost, Tolin, Steketee, Fitch, & Selbo-Bruns, 2009; Mataix-Cols et al., 2013). People with HD attribute the need to save possessions to a strong emotional attachment and distorted beliefs about possessions, such as their importance (e.g. “I will never be able to replace this”), their responsibility for the items (“I am responsible for finding a use for this item”), and the need for control over items (“No-one has the right to touch my possessions”) (Steketee, Frost, & Kyrios, 2003). In addition to the strong negative emotions experienced when attempting to discard items, people with HD report strong positive emotions and comfort from acquiring or rediscovering possessions, which also reinforces the saving behaviour (Frost & Hartl, 1996; Frost, 2010).

HD can have a considerable negative impact on quality of life (Saxena et al., 2011). People with HD often have poor physical health, an increased likelihood of earning an income below the poverty line, and of being in debt and/or unemployed compared to controls (Nordsletten, Reichenberg, Hatch, Fernández De La Cruz, et

al., 2013; Tolin, Frost, Steketee, Gray, et al., 2008). They are also more likely to be unmarried, live alone, and have more problems with relationships and family functioning (Frost, Steketee, & Williams, 2000; Mataix-Cols et al., 2013; Tolin, Frost, Steketee, & Fitch, 2008). A qualitative study by Kellett and colleagues (Kellett, Greenhalgh, Beail, & Ridgway, 2010) found that people with HD experience feelings of shame, embarrassment, and stigma in relation to their hoarding. HD is also highly comorbid with other psychiatric conditions; between 50-69% have at least one other comorbid psychiatric condition such as anxiety or depression (Frost, Steketee, et al., 2011; Mataix-Cols et al., 2013).

1.2 Characteristics of HD

The onset of hoarding behaviour is typically observed in early to mid-adolescence and the illness course is often long-term, with more severe symptomatology emerging in later years (Ayers, Saxena, Golshan, & Wetherell, 2010; Grisham, Frost, Steketee, Kim, & Hood, 2006; Ivanov et al., 2013; Seedat & Stein, 2002; Tolin, Meunier, Frost, & Steketee, 2010). HD seems to be equally prevalent across genders (Nordsletten, Reichenberg, Hatch, Fernández De La Cruz, et al., 2013; Timpano, Exner, et al., 2011), although some studies have reported higher rates in men (Iervolino et al., 2009; Samuels et al., 2008).

Most research into HD has been conducted in the UK or the USA (e.g. Frost, 2010; Mataix-Cols et al., 2013; Steketee & Frost, 2003), with similar prevalence rates and clinical presentation reported in German studies (Mueller et al., 2009; Timpano et al., 2013). There is currently little research in other cultural groups across the globe. Matsunaga and colleagues (Matsunaga, Hayashida, Kiriike, Nagata, & Stein, 2010) found hoarding symptoms in a Japanese OCD sample and recently Timpano et al. (2015) have studied hoarding symptoms in student samples in China.

A common characteristic of HD is the failure to recognise hoarding as problematic. Tolin, Fitch, Frost, and Steketee (2010) found that over 50% of family members rated their hoarding relative as having poor or no insight into their condition. Furthermore, HD treatment studies have found high rates of treatment refusal and drop out, with proposed links between lack of insight and motivation for treatment (Frost et al., 2010).

A widely noted correlate of HD is neurocognitive difficulties. A recent review by Woody, Kellman-McFarlane, and Welsted (2014) described replicable difficulties in people with HD compared to controls in areas of problem solving, planning, organisation, working memory/sustained attention, and visuospatial learning and memory. People with HD also report poorer confidence in their memory (Hartl et al., 2004). Some studies have identified symptoms of attention deficit disorder in people with HD (e.g. Frost, Steketee, et al., 2011). Problems with attention contribute to some of the clinical features of the disorder, such as difficulty focusing on organising possessions (Hartl et al., 2005; Tolin & Villavicencio, 2011).

Indecisiveness is another manifestation of the cognitive differences present in HD. Problems with indecisiveness are positively correlated with increased hoarding severity (Frost, Tolin, Steketee, & Oh, 2011) and are elevated in HD compared to OCD groups (Grisham, Norberg, Williams, Certoma, & Kadib, 2010; Samuels et al., 2007). Neuroimaging studies have supported this finding. For example, Tolin and colleagues found that during decision making tasks, people with HD display unusual activity on functional magnetic resonance imaging, including hyperactivity in the fronto-temporal lobes and abnormal haemodynamic activity when compared with healthy controls (Tolin, Stevens, Nave, Villavicencio, & Morrison, 2012; Tolin, Stevens, Villavicencio, et al., 2012). Preliminary evidence shows that abnormal haemodynamic activity normalises following cognitive behavioural therapy for HD (Tolin, Stevens, Nave, et al., 2012).

1.3 Causal explanations

Various causal explanations for HD have been suggested in the literature, including genetic and psychosocial factors.

Genetic research suggests that the symptoms of HD are at least partially attributable to heritable factors (Iervolino et al., 2009; Mathews et al., 2007; Samuels et al., 2007). In one of the earlier studies, comprising 32 participants who self-identified as 'chronic savers and pack rats', the majority (78%) reported also having a first-degree relative with these problems (Frost & Gross, 1993). Data drawn from a study of 2,053 monozygotic and dizygotic twins found that approximately 50% of the variance in hoarding symptoms was accounted for by genetic factors (Iervolino et al.,

2009). The remaining variance was attributable to non-shared environmental factors and measurement error.

Several studies have found a higher incidence of traumatic and stressful life events in people who hoard compared to controls or OCD patients (Cromer, Schmidt, & Murphy, 2007; Grisham et al., 2006; Hartl et al., 2005; Landau et al., 2011; Timpano, Keough, Traeger, & Schmidt, 2011) and up to 55% of people report that the onset or worsening of their HD was associated with a stressful or traumatic event (Ayers et al., 2010; Grisham et al., 2006; Landau et al., 2011). Particular types of stressful life events which appear to be associated with HD are possession related events (e.g. forcible removal of possession by parent; Landau et al., 2011; Hartl et al., 2005) and loss of or significant changes to relationships (e.g. divorce, death) (Tolin et al., 2010). Contrary to lay belief, HD does not seem to be associated with material deprivation (Frost & Gross, 1993; Landau et al., 2011). A recent study by Medard and Kellett (2014) indicated that people with HD have higher rates of attachment difficulties (attachment avoidance and attachment anxiety) than control groups, which may play a role in the aetiology and/or maintenance of hoarding behaviour.

1.4 Wider impact of HD

The problems linked to HD in the wider family system have been increasingly recognised over the past few years. Evidence suggests high levels of family burden (Drury, Ajmi, Fernández de la Cruz, Nordsletten, & Mataix-Cols, 2014), negative impact on quality of relationships, and increased family conflict are positively linked to HD (Sampson, 2013; Wilbram, Kellett, & Beail, 2008). Family members also report stigma and embarrassment about the hoarding (Wilbram et al., 2008) and higher levels of childhood distress if they grew up in a home with someone who hoarded (Tolin, Frost, Steketee, & Fitch, 2008). Drury et al. (2014) found that functional impairment in relatives was equivalent to that experienced by the individuals with HD themselves. Lack of insight and willingness to seek help can be a major source of frustration and distress for family members. Tolin et al. (2008) found that levels of family rejection (i.e. frustration and hostility) were higher if the hoarding relative had lower levels of insight and more severe hoarding symptoms.

The consequences of HD extend beyond the individual and their family. Large amounts of clutter in the home can lead to unsanitary and even dangerous living conditions for people with HD, their families, and neighbours; one survey of health departments in the USA reported that 67% of cases of hoarding were judged to be a serious fire hazard (Frost, Steketee, & Williams, 2000). Significant social and economic costs are typically associated with HD (Tolin, Frost, Steketee, Gray, & Fitch, 2008). The financial costs are linked to missed days of work, the poorer physical health of people with HD necessitating greater use of health services, and increased use of social services related to addressing housing and childcare concerns (Tolin, Frost, Steketee, Gray, et al., 2008). The cost of house clear-outs can also be substantial, with one health department in the USA reporting to have spent £10,700 on a single clear-out (Frost et al., 2000).

1.5 Cognitive behavioural model of HD

Frost and Hartl (1996) proposed a cognitive-behavioural model of 'compulsive hoarding' based on early research and clinical experience. They defined compulsive hoarding as: (1) the acquisition of, and failure to discard a large number of possessions that appear to be useless or of limited value; (2) living spaces sufficiently cluttered so as to preclude activities for which those spaces were designed; and (3) significant distress or impairment in functioning caused by the hoarding. This definition was highly influential in the creation of the DSM-5 diagnostic criteria (Mataix-Cols et al., 2010).

The model proposed that people with HD have information processing deficits (in decision making, categorisation/organisation and memory functioning) which are linked to certain beliefs and behaviours, resulting in the saving behaviour seen in HD. According to the model, people with HD will hold erroneous beliefs about possessions and the negative consequences of discarding (i.e., a fear of making mistakes and an over-focus on the negative costs of discarding). These beliefs, coupled with the deficits in information processing mean that discarding is avoided. The model also suggests an unusual level of emotional attachment to possessions which has been confirmed in many empirical studies (e.g. Frost, Hartl, Christian, & Williams, 1995; Grisham et al., 2009; Nedelisky & Steele, 2009).

In summary, HD is proposed to be a disorder of dysfunctional beliefs and emotions associated with the acquisition and discarding of possessions, resulting in behavioural avoidance and a build-up of clutter. The repercussions of the disorder are wide-reaching for the individual, their family, and society. The causes suggested by the research are a combination of bio-psycho-social factors.

1.6 The development of treatments for HD

The poor response of hoarding symptoms to standard OCD treatment (see meta-analysis by Bloch et al., 2014) prompted the development of treatments specifically for hoarding problems. The first study to pilot cognitive-behavioural therapy (CBT) for hoarding based on Frost and Hartl's (1996) model was conducted by Hartl and Frost (1999). The authors presented a case study of a 53 year old woman with compulsive hoarding who engaged in CBT treatment over 17 months targeting the maintaining factors suggested by the model, including training in decision-making and categorization, exposure to discarding, and cognitive restructuring of her dysfunctional beliefs related to hoarding. They found substantial improvements in clutter, hoarding symptoms, and indecisiveness after the intervention. Steketee and colleagues (Steketee, Frost, Wincze, Greene, & Douglass, 2000) replicated the positive outcomes on hoarding symptoms in a pilot trial of 7 hoarding participants, 6 of whom completed group-based CBT and 1 individual CBT. Two further single case experimental design studies have tested variants of the CBT for HD protocol (Kellett, 2006; Pollock, Kellett & Totterdell, 2014) with promising results in terms of reduction in clutter and hoarding symptoms.

A number of different CBT for HD protocols for individual (Steketee & Frost, 2007, 2013), group (Muroff, Underwood, & Steketee, 2014), and self-help treatment have been developed (Bubrick, Neziroglu, & Yaryura-Tobias, 2004; Tolin, Frost, & Steketee, 2007b). Typically, CBT for HD includes motivational interviewing techniques (Miller & Rollnick, 2002) to enhance engagement in treatment, psychoeducation about HD, creation of an individualised formulation of hoarding behaviour, skills training in organising, sustaining attention and making decisions about belongings, exposure to discarding and not acquiring new possessions, and evaluation and cognitive restructuring of dysfunctional beliefs about possessions

(e.g. Steketee & Frost, 2013). Treatment often involves home visits for 'in vivo' exposure and skills teaching. More recently, approaches other than CBT have been developed to treat HD, such as 'harm reduction' for clients not motivated to change their behaviour (Tompkins, 2015), pharmacological treatments using serotonin reuptake inhibitors (SRIs; Saxena, Brody, Maidment, & Baxter Jr., 2007; Saxena & Sumner, 2014), interventions utilising cognitive remediation to address neurocognitive deficits (Ayers et al., 2014; Dimauro, Genova, Tolin, & Kurtz, 2014), and family-based interventions to enhance motivation of the person with HD and/ or support the needs of family members (Chasson, Carpenter, Ewing, Gibby, & Lee, 2014; Sampson, Yeats, & Harris, 2012; Tompkins, 2011).

Despite the increasing amount of research in this area, evidence-based treatments for HD are still in their infancy. Progress thus far may have been limited in part by the challenges posed by some of the features typical of HD, including: poor insight and motivation for seeking treatment, psychiatric comorbidity, cognitive difficulties including memory and attention, and the long length of treatments which often include home visits, which make treatments expensive and resource heavy. Even once treatment has begun, numerous challenges remain including resistance to change from individuals with HD, avoidance of discarding, the presence of strong beliefs developed over many years regarding possessions, and potential defensiveness developed through years of exposure to other's misunderstanding of their hoarding as 'messiness' or 'laziness' (Frost et al., 2010; Tolin, 2011). Additionally, many of the treatment trials were conducted before the DSM-5 criteria were published and thus may have included participants who would not meet full criteria for HD using current guidance, or whose hoarding symptoms were attributable to other disorders.

1.7 Previous reviews of treatments for HD

Two treatment reviews have been conducted to date which examined the outcomes of psychological and pharmacological treatments for hoarding difficulties (Muroff, Bratiotis, & Steketee, 2011 and Tolin, Frost, Steketee, & Muroff, 2015). Muroff et al.'s (2011) review included treatments for OCD such as exposure and response prevention, as well as HD specific treatments. They found a generally poor response

and higher dropout rates for non-specific treatments in patients with hoarding symptoms, compared to those without. Improved results were found for trials which used hoarding-specific treatments including individual CBT (Steketee, Frost, Tolin, Rasmussen, & Brown, 2010; Tolin, Frost, & Steketee, 2007a; Turner, Steketee, & Nauth, 2010); group CBT (Gilliam et al., 2009; Muroff, Steketee, & Bratiotis, 2010; Muroff et al., 2009; Pekareva-Kochergina & Frost, 2009), internet delivered interventions (Muroff, Steketee, & Frost, 2009; Muroff, Steketee, Himle, & Frost, 2010), and one prospective pharmacotherapy study (Saxena, Brody, Maidment, & Baxter Jr., 2007).

Tolin et al.'s (2015) meta-analysis found a significant reduction in overall hoarding severity after CBT for HD, with large effect sizes (Hedges g=0.82). The authors divided hoarding symptoms into difficulty discarding, clutter, and acquiring. They found the strongest effect for difficulty discarding, the primary symptom of HD. However, scores at the end of treatment remained mostly in the clinical range, with only 35% of participants achieving clinically significant change on overall HD severity. A smaller effect size was found for improvements in functional impairment caused by the HD (Hedges g=0.52; moderate effect size). Tolin and colleagues also investigated moderators of CBT outcomes; female gender, younger age, taking psychiatric medications, a greater number of CBT sessions and more home visits were all associated with better outcomes. Whether the intervention was in group or individual format was not predictive of outcome.

Though the reviews by Muroff et al. (2011) and Tolin et al. (2015) extended our understanding of HD treatments, some important limitations were noted with regard to the scope of the reviews and adopted methodology. Muroff et al.'s (2011) review was completed prior to the creation of diagnostic criteria for HD and included studies of people with OCD plus 'hoarding behaviours', who may comprise a distinct group of patients to those with 'pure' HD who are likely to respond differently to treatment (Pertusa, Frost, & Mataix-Cols, 2010). In addition, the authors did not limit their studies to people meeting criteria for 'compulsive hoarding' (e.g. from Frost & Hartl, 1996), or the proposed DSM-5 criteria, meaning that patients may not have met full criteria for HD. The inclusion of studies in the review using non-HD specific

outcome measures such as the Y-BOCS (Goodman, 1989) makes the interpretation of results in terms of improvement in HD symptoms difficult and is an important limitation. Tolin et al.'s (2015) meta-analysis had a more defined focus, but opted to review CBT-based interventions only, thus rendering questions regarding alternative treatments unanswered. Given the multifaceted nature of HD, such as the impact on family members and comorbid psychiatric conditions, improving our understanding of a broader range of evaluated treatments that include, but are not limited to, CBT would have merit.

In terms of limitations around methodology, Muroff et al.'s choice of a narrative review is more prone to error and bias than a systematic review (Cook, 1997) and does not include an assessment of the quality of the studies. Furthermore, the meta-analytical methodology adopted by Tolin et al. (2015) is not without limitations in terms of how best to summarise the data, particularly due to the heterogeneous study designs (open trials vs randomised control trials) and intervention formats (individual CBT, group CBT, non-professionally facilitated support groups). Additionally, the Tolin meta-analysis did not investigate longer term effects of interventions (i.e. follow-up assessment results) and many of the studies reviewed were not RCTs, limiting our interpretation of effect sizes which are often inflated in uncontrolled studies.

1.8 The current review

Research into treatments for HD has been limited in part by the previous lack of diagnostic criteria and the intrinsic challenges of the disorder itself (e.g. low levels of treatment seeking). Since the publication of the DSM-5 criteria, there has not been a review of the diverse developing treatments for HD. Given the aforementioned gaps left by the two published treatment reviews, a systematic review of treatments for HD is warranted which uses stringent methodology, includes approaches to treatment other than CBT and which will report more thoroughly on the risk of bias in the existing treatment studies. The present study aims to systematically review treatment interventions for hoarding, including outcomes for hoarding symptoms, associated impairment, comorbid emotional difficulties, and impact on family members. An assessment of the methodological rigour of the studies will be

undertaken, which previous reviews have not done. The results should support clinicians in choosing and delivering evidence based treatments for HD, and identifying future targets for research.

The systematic review will investigate whether treatments for people with HD or their families demonstrated:

i. reductions in severity of hoarding symptoms (primary outcome); and
ii. reductions in comorbid psychiatric symptoms (depression, anxiety), or quality of life (secondary outcomes).

2. Method

2.1 Search strategy

Systematic searches were performed to find relevant treatment studies from the following databases: PsycINFO (1806 to January 2015), MEDLINE (R) (1946 to January 2015), EMBASE (1974 to January 2015), and Web of Science. Databases were searched in January 2015. The search strategy was as following: "treatment" OR "intervention" OR "psychological intervention*" OR "therap*" OR "psychotherapy*" OR "CBT" OR "ERP" OR "medication" OR "psychosocial" OR "bibliotherap*" OR "pharmacotherapy*" OR "drug therap*" OR "support group" OR "cognitive therap*" OR "behavio*r therap*" OR "cognitive behavio*r therap*" OR "motivational interview*" OR "family therap*" OR "self-help" OR "psychoeducation*" OR "harm reduction" AND "Hoarding disorder" OR "compulsive hoarding" OR "hoarding behavio*r" OR "clutter" OR "obsessive hoarding" OR "obsessive compulsive hoarding" OR "hoard*".

Searches were limited to human studies in the PsycINFO, MEDLINE, and EMBASE databases, and excluded after searches for Web of Science. The explosion function was not used, due to hoarding being a relatively new subject heading term. To identify as many articles as possible, we did not limit the search to English only in the initial stages of the search.

Duplicates were removed, and titles and abstracts then manually screened for eligibility. Papers that did not meet the inclusion criteria were rejected. Full text articles were retrieved for potentially eligible studies, and the reference lists of final

selected studies and relevant review articles were searched manually for additional studies.

2.2 Inclusion and exclusion criteria:

Papers were included in the review if:

(i) the study evaluated an intervention for individuals or relatives of individuals with HD (according to the working criteria prior to the publication of the DSM-5 (Mataix-Cols et al., 2013) or the final DSM-5 diagnostic criteria (APA, 2013)), or 'compulsive hoarding' (as defined by Frost and Hartl (1996) or the presence of clinically significant hoarding symptoms defined according to standardized measures);

(ii) outcome measures were reported (including measures of hoarding symptoms or impact on life/distress, co-morbid psychiatric symptoms); and

(iii) the paper was published in an indexed journal or published abstract from a professional/research conference.

Studies were excluded from the review if:

(i) the paper was a case study, single case experimental design (n=1) or a discussion article;

(ii) the paper was not available translated into English;

(iii) the study reported on animal hoarding only (given animal hoarding is thought to form a distinct issue (Frost, Patronek, & Rosenfield, 2011));

(iv) the paper was an unpublished thesis or dissertation.

The first author (CT) assessed the relevance of articles against above criteria, and queries were resolved through discussion with author LFC.

2.3 Data extraction

A data extraction form was developed for the purpose of the review, based on the Cochrane Collaboration's data extraction template (2011) and the quality assessment tool. An example of the data extraction form can be found in Appendix

2. CT conducted the data extraction. Where partial eta squared were reported for effect sizes, these were converted into Cohen's d (Cohen, 1988) using a formula from DeCoster (2012).

2.4 Quality assessment

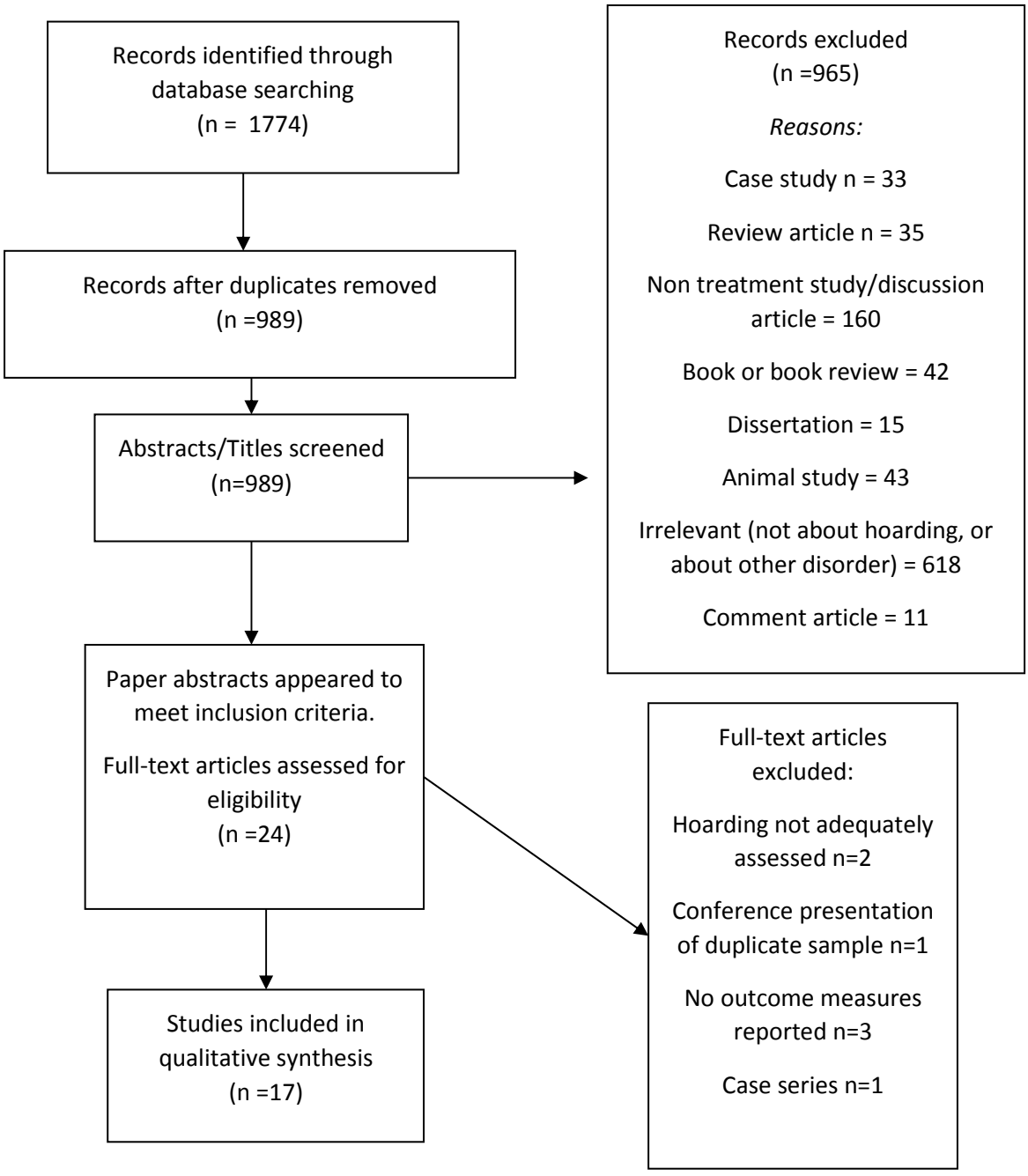
The quality of each selected paper was assessed using the Clinical Trials Assessment Measure (CTAM, Tarrier & Wykes, 2004; see Appendix 3). Alternative quality assessment tools were considered (e.g. Effect Public Health Practice Project's Quality Assessment Tool for Quantitative Studies), but the CTAM eventually selected as it was designed specifically to assess the methodological quality of psychological treatments (which made up the majority of papers selected) (Tarrier & Wykes, 2004; Wykes, Steel, Everitt, & Tarrier, 2008), covered a range of areas of potential bias relevant to psychological treatments (e.g. blinding) and provides a numerical score which allows comparison of study quality to other systematic reviews of treatments which have used the CTAM. The CTAM includes 6 subscales for assessment including items to assess the risk of bias in studies: recruitment method and sample size; allocation to treatment (randomisation process); assessment procedures; control groups; description of treatment; and analysis. The CTAM has a maximum possible score of 100 and Wykes et al. (2008) recommend a score of 65 and above as demonstrating adequate methodology. It has good inter-rater reliability and concurrent validity (Tarrier & Wykes, 2004). Two raters (CT and LFC) blind-rated selected studies and any disagreements were resolved through discussion.

3. Results

3.1 Search results

The search identified 989 papers after duplicates were removed. Figure 1 shows the process of the systematic search. No additional studies were identified through hand searching reference lists. A total of 17 papers were identified for review, comprising 17 distinct samples. The Frost, Pekareva-Kochergina, and Maxner (2011) paper included two separate studies (study 1 and study 2) and Muroff, Steketee, Frost, and Tolin (2014) was a 1 year follow-up of Steketee, Frost, Tolin, Rasmussen and Brown (2010).

Figure 1: PRISMA (2009) flow diagram of systematic review process



3.2 Sample characteristics

The studies included and participant characteristics are described in table 1 below. All studies were conducted in the USA. Five studies used the DSM-5 diagnostic criteria for HD (3 studies) or the working criteria prior to publication (2 studies). The remainder used Frost and Hartl's (1996) definition of 'compulsive hoarding' (3 studies), or defined clinically significant hoarding symptoms according to standardized measures (9 studies). Sixteen studies looked at the efficacy of treatment for people with HD/compulsive hoarding, and one was an intervention for

relatives only (Chasson et al., 2014). The seventeen studies contained a total of 474 patients and 9 relatives of patients with HD/'compulsive hoarding'. The number of participants with HD in each study ranged from 6 to 115 (mean=28.41, SD=26.21, median=17), with an average dropout of 14.3% from the 15 studies which reported dropout rates. Most studies had small sample sizes, with only eight studies including more than 20 participants.

Participants in the studies were reported as being mainly white/Caucasian ethnic background, female, and middle-aged (m=56.63, SD=7.84). Three studies were aimed specifically at older people (range 56-87) with HD (Ayers et al., 2014; Ayers, Loebach, Golshan, & Saxena, 2011; Turner et al., 2010) and the remainder included adults of any age.

3.3 Design

Most studies used a pre-post design with no control group (12/17), 3 used a wait-list control, and 2 had active control groups; one used relaxation training as the control intervention and the other used self-help CBT bibliotherapy for HD (see table 1). The two pharmacological studies were open label. Four out of the five controlled trials randomised participants to conditions, and the other was quasi-experimental as it used an existing wait-list control. Four studies were longitudinal and included 1-month (Frost et al., 2011, studies 1 & 2), 6-month (Ayers et al., 2011), or one year (Muroff et al., 2014) follow-up measurement points.

Table 1: Characteristics of studies included in the review

Study	N (started treatment/ allocated)	Dropout	Mean age (SD)	Gender (% female)	Ethnicity	Intervention type	No. of sessions	Study design (control group)	HD measures used	Primary outcome	Assessment period	How HD established
Ayers et al. (2011)	12	0 (2 lost at FU)	73.66 (6.54)	58.3%	83.33% white 16.67% Hispanic	Individual CBT	26	Uncontrolled trial	UHSS, SI-R, CIR	HD Sx	Pre, post, 6 month FU	Proposed DSM-5 criteria
Ayers et al. (2014)	11	0	66 (7)	81.8%	90.9% white 9% Hispanic	Cognitive remediation + behavioural therapy	24	Uncontrolled trial	UHSS, SI-R, CIR	HD Sx	Pre, mid, post	DSM-5 criteria
Chasson et al. (2014)	9	4 (44%)	46.22 (13.10)	77.8%	88.89% white 11.11% Asian American	Family-as-motivators training	14	Uncontrolled trial	HRS	Family wellbeing	Pre, mid, post	HRS scores in clinical range

Dimauro et al. (2014)	17	0	60 (13)	77%	100% white	Cognitive remediation	24	RCT (Relaxation training)	SI-R	Cognitive functioning	Pre, mid, post	HRS + interview approximated to DSM-5 criteria
Frost et al. (2011) Study 1	17	0 (2 did not complete post measures but attended FU)	53.7 (9.8)	88.2%	100% white	Biblio-based support group	13	Uncontrolled trial	SI-R, SCI, CIR	HD Sx	Pre, mid, post, 1 month FU	SI-R cut-off
Frost et al. (2011) Study 2	12 (10 above cut off on HRS/SIR)	1 (8.3%)	61.3 (9.3)	75%	100% white	Biblio-based support group	13	Uncontrolled trial	SI-R, SCI. HRS and CIR pre-post only	HD Sx	Pre, mid, post, 1 month FU	SI-R and HRS cut-off
Frost et al. (2012)	43	5 (12%)	57 (11)	WL= 90% Tx= 94.4%	WL= 90% white TX =100% white	Biblio-based support group	13	RCT (Wait list)	SI-R, SCI, CIR, HRS	HD Sx	Pre, post	SI-R and HRS cut-off
Gilliam et al. (2011)	45	15 (33%)	55.06 (10.63)	86.7% F	88.8% white	Group CBT	16-20 (m=17.6)	Uncontrolled trial	SI-R	HD Sx	Pre, mid, post	SI-R + interview
Muroff et al. (2009)	32	Not reported	53	65.6% F	90.6% white	Group CBT+2 individual sessions	16-20 (m=16.6)	Uncontrolled trial	SI-R, CIR, SCI	HD Sx	Pre, post	HRS + interview

Muroff et al. (2010)	115	Not reported (30 completed all measures)	LT =54 (8.17) ST= 49.73 (9.29) WL =49.5 (9.21)	LT =97.6% ST =92.7% WL =94.4%	WL=94.4 % white ST =94.5% white LT= 92.5% white	Online group CBT/self help	n/a	Quasi-experimental wait-list controlled trial	SI-R, CIR	HD Sx	Pre, every 3 months over 1 year	Self-identified as meeting Frost & Hartl (1996) definition
Muroff et al. (2012)	39	1 (2%)	56.87 (8.12)	76.3%	84% white 15.8% black 2.6% Native American , 2.6% Hispanic	Group CBT with or without additional HA	20 + 4 HA	RCT (Biblio-therapy)	HRS, SI-R	HD Sx	Pre, mid, post	HRS + interview
Saxena et al. (2007)	32	7 (22%)	48.7 (9.7)	71.8%	not reported	Paroxetine up to 60mg/day	10-12 weeks	Uncontrolled trial	YBOCS, subset UCLA-HSS	HD Sx	Pre, post	Frost & Hartl (1996) definition
Saxena & Sumner (2014)	24	1 (4%)	51.8 (8.1)	87.5%	79.17% white 12.5% Hispanic 2 8.3% Asian American	Venlafaxine extended release up to 300mg/day	12 weeks	Uncontrolled trial	UHSS, SI-R	HD Sx	Pre, post	DSM-5 criteria

Steketee et al. (2010) and Muroff et al. (2014)	44	7 (16%)	54	75%	87% white, 2% Latino 10% Black, 2% Asian.	Individual CBT	26	RCT (Wait list, up to session 12)	SI-R, HRS	HD Sx	Pre, mid (session 12), post, 1 year FU	HRS + interview
Tolin et al. (2007a)	14	4 (29%)	49.2 (14.96)	100%	80% white 10% African American 10% Hispanic	Individual CBT	26	Uncontrolled trial	CIR, SI-R	HD Sx	Pre, mid, post	Frost & Hartl (1996) definition
Tolin et al. (2012)	6	0	52.83 (6.71)	33%	83.3% White	Group CBT	16	Uncontrolled trial	SI-R, HRS	Brain functioning/HD Sx	Pre, post	Proposed DSM-5 criteria
Turner et al. (2010)	11	5 (45%)	72.3	83.3 (compl-eters)	not reported	Individual CBT	28-41 (m=35.3)	Uncontrolled trial	CIR, first 3 items HRS	HD Sx	Pre, post	CIR cut-off + difficulty discarding

HD Sx=Hoarding Disorder Symptoms, FU=Follow-up, LT=Long-term members, ST=Short-term members, WL=Wait-list, RCT=Randomised Control Trial, HA=Home Assistance visits. Measures: CIR=Clutter Image Rating scale, HRS=Hoarding Rating Scale, SI-R=Saving Inventory Revised, SCI=Saving Cognitions Inventory, UHSS=UCLA Hoarding Severity Scale, YBOCS=Yale-Brown Obsessive Compulsive Scale

3.4 Description of interventions used

The treatment interventions employed were as follows: individual CBT (4 studies), group CBT (4 studies), CBT-based bibliotherapy support groups (3 studies), cognitive remediation (2 studies), pharmacotherapy (2 studies), 1 online support group, and 1 'family-as-motivators' training.

All studies which trialled individual CBT (e.g. Ayers, Loebach, Golshan, & Saxena, 2011; Muroff et al., 2014; Steketee, Frost, Tolin, Rasmussen, & Brown, 2010; Tolin, Frost, & Steketee, 2007; Turner, Steketee, & Nauth, 2010) based their intervention on Steketee and Frost's (2007) manual of CBT for compulsive hoarding. This protocol recommends 26 weekly sessions of CBT, including in-home sessions. Ayers et al. (2011) increased the intensity of the intervention to twice weekly for the first 20 sessions and Turner et al. (2010) delivered additional treatment sessions to the proposed number reported in the original protocol (m=35.3, range 28-41).

The four studies using group CBT (Gilliam et al., 2011; Muroff, Steketee, Bratiotis, & Ross, 2012; Muroff, Steketee, Rasmussen, et al., 2009; Tolin, Stevens, Nave, et al., 2012) also used the Steketee and Frost (2007) manual, but adapted it for a group setting and delivered fewer sessions (range=16-24). Muroff et al. (2009) also offered 2 home visits to participants, and their later study (Muroff et al., 2012) included 4 'home assistance' (HA) visits in one experimental condition. The bibliotherapy CBT-based group studies (Frost et al., 2011, studies 1 and 2; Frost, Ruby, & Shuer, 2012) consisted of 13 sessions and followed Tolin, Frost, and Steketee's (2007b) CBT-based self-help book "*Buried in treasures*". All were facilitated by non-professionals. The bibliotherapy control condition of Muroff et al.'s (2012) study also used Tolin et al.'s (2007b) book. The online support group (Muroff, Steketee, Himle, et al., 2010) used CBT methods from Steketee and Frost (2007)'s manual plus peer support.

Ayers et al. (2014) created an idiosyncratic protocol consisting of cognitive remediation techniques from Twamley, Vella, Burton, Heaton, and Jeste's (2012) protocol plus exposure to discarding and not acquiring (a component of CBT). The second cognitive remediation trial (Dimauro et al., 2014) used a computerized program ("Scientific Brain Training PRO") which targeted specific cognitive functions

associated with HD including attention and memory, but did not directly target hoarding symptoms.

In terms of the two pharmacotherapy trials (Saxena, Brody, Maidment, & Baxter Jr., 2007; Saxena & Sumner, 2014), Paroxetine (a selective serotonin reuptake inhibitor, SSRI) and Venlafaxine (a serotonin–norepinephrine reuptake inhibitor, SNRI) were used, following a protocol of incremental increases in dosage.

In the final study, which was designed for relatives (Chasson et al., 2014), the 'family-as-motivators' intervention comprised different components including psychoeducation about HD, harm reduction techniques, training in motivational interviewing, and training in reducing family accommodation of HD, and was based on an unpublished manual (Chasson, Ewing, Gibby, & Carpenter, 2013).

3.5 Outcome measures used

All included studies used at least one published, validated measure of hoarding symptoms (primary outcome). Measures of general hoarding symptomatology (i.e. difficulty discarding, clutter, and impairment and/or distress) included: Hoarding Rating Scale (HRS; Tolin, Frost, & Steketee, 2010), Saving Inventory-Revised Scale (SI-R; Frost, Steketee, & Grisham, 2004), UCLA Hoarding Severity Scale (UHSS; Saxena, Brody, Maidment, & Baxter Jr., 2007), and Clutter Image Rating scale (CIR; Frost, Steketee, Tolin, & Renaud, 2008). The Saving Cognitions Inventory (SCI; Steketee, Frost, & Kyrios, 2003) was also included, which measures beliefs about possessions (e.g. emotional attachments, responsibility for items). For the purposes of the qualitative synthesis we looked only at total scores on these questionnaires, due to heterogeneity in reporting of the individual symptom dimensions and the variety of outcome measures used.

Secondary outcome measures also varied between the studies. Measures of anxiety and depression symptoms were: Beck Depression Inventory (BDI; Beck, Steer, & Brown, 1996; Beck, Ward, Mendelson, Mock, & Erbaugh, 1961), Beck Anxiety Inventory (BAI; Steer & Beck, 1997), Depression Anxiety Stress Scale (DASS; Lovibond & Lovibond, 1993), Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983), Hamilton Depression Rating Scale (HDRS; Hamilton, 1960b), Hamilton Anxiety Rating Scale (HAM-A; Hamilton, 1960a), and the Y-BOCS (Goodman, 1989). Measures

of the impact of hoarding on daily living/functional impairment included: Activities of Daily Living Scale – Hoarding (ADL-H; Frost, Hristova, Steketee, & Tolin, 2013; Grisham, Frost, Steketee, Kim, & Hood, 2006), Sheehan Disability Scale (SDS; Sheehan, 1983), and the Quality of Life Scale (Burckhardt, Woods, Schultz, & Ziebarth, 1989). The intervention study with relatives also included specific measures of the impact of HD on family members, including the Family Member Impact Scale (Orford, Templeton, Velleman, & Copello, 2005) and the Family Response to Hoarding Scale (Steketee & Frost, 2013a).

3.6 Quality assessment

The CTAM (Tarrier & Wykes, 2004) was used to rate the methodological quality of each study from 0-100. The total scores are presented in Table 2 below. As can be seen, none of the studies scored above the suggested cut off of 65 for adequate quality suggested by Wykes et al., (2008). It should be noted however, that the CTAM has generally been used with randomised control trials (RCTs) and therefore this cut-off may not effectively reflect the specifics of the uncontrolled studies. The average score on the CTAM was 29.58 (SD=15.37, range 16-64, median=24). The average score of the RCTs was higher, whereas the uncontrolled trials were generally of lower quality.

Table 2: Scores on the CTAM

RCTs (First author, date)	CTAM total score	Grand mean (SD)
DiMauro (2014)	64	
Frost (2012)	34	
Muroff (2012)	63	
Steketee (2010)	39	
		50 (15.73)
Uncontrolled trials (First author, date)		
Ayers (2011)	26	
Ayers (2014)	46	
Chasson (2014)	20	
Frost Study 1 (2011)	21	
Frost Study 2 (2011)	21	
Gilliam (2011)	25	
Muroff (2009)	24	
Muroff (2010)	19	
Saxena (2007)	19	
Saxena (2014)	18	
Tolin (2007)	17	
Tolin (2012)	31	
Turner (2010)	16	
		23.3 (17.98)

Mean scores in each CTAM domain are shown in table 2 (see Appendix 5). Most studies did not score highly in any domains of the CTAM. Items which contributed to lower scores were: sampling, allocation, assessment methodology, and lack of control groups. Participant samples were usually made up of volunteers from advertisements, which attracted a rating of zero on the CTAM. Only 6 studies scored on this item (Chasson et al., 2014; Gilliam et al., 2011; Muroff et al., 2009, 2012; Steketee et al., 2010; Turner et al., 2010). Sample sizes were generally small – only 4/17 studies had more than 27 participants per treatment group (Gilliam et al., 2011; Muroff et al., 2010; Muroff et al., 2009; Saxena et al., 2007), and the RCTs also lost points on this item. No studies reported whether a power analysis was conducted to determine the appropriate sample size for the trial. Another domain in which most studies scored poorly was allocation. Only 4 studies randomised subjects (Dimauro et al., 2014; Frost, Ruby, & Shuer, 2012; Muroff et al., 2012; Steketee et al., 2010) and,

of these, only one paper provided details about the randomisation process (Frost et al., 2012). No studies described that randomisation was independent of the research team.

In terms of assessment, all studies scored full marks on the use of standardised assessment measures. However, only 3/17 studies reported to have used independent and blind assessors (Ayers et al., 2011; Dimauro et al., 2014; Muroff et al., 2012), and one further study used independent assessors (Tolin et al., 2012). No studies described the blinding procedure or verified blinding. Only 5 of the 17 studies had a control group, and only 2/17 studies had an active control group which scored on the CTAM (Dimauro et al., 2014; Muroff et al., 2012). All studies used appropriate analysis techniques, but some lost marks on their reporting or handling of drop-outs. 9/17 studies described appropriate handling of dropout or had less than 15% attrition. Two studies scored full marks on the analysis questions and both were RCTs (DiMauro et al., 2014; Steketee et al., 2010). On the treatment section, all studies used either a treatment manual or described the treatment well, but only 2/17 rated adherence to treatment protocol (Ayers et al., 2011; 2014). Ayers et al. (2011) checked for both fidelity to the protocol and treatment competence, and Ayers et al. (2014) checked for fidelity only. It should be noted that the items on adherence to protocol of use of manuals were less relevant to the two pharmacological studies (Saxena & Sumner 2014; Saxena et al., 2007).

3.7 HD Outcomes by type of intervention

Table 3 (see appendix 6) presents an overview of which studies found significant improvements in HD symptoms, associated functional impairment, and comorbid anxiety and depression. Across all studies, outcomes were considered significant at the $p<0.05$ level for the purposes of the synthesis.

3.7.1 CBT interventions

Individual CBT

All individual CBT studies reported significant improvements in hoarding symptoms on at least one measure by the end of treatment. Only one study (Ayers et al., 2011) used proposed DSM-5 criteria, whereas the rest of the individual CBT studies used either Frost and Hartl's (1996) criteria or questionnaire cut-offs. Three studies (Ayers

et al., 2011; Steketee et al., 2010; Tolin et al., 2007a) reported significant improvement on the SI-R (Frost et al., 2004) of between 15-28% reduction in scores at post-treatment, with large effect sizes (ES) of Cohen's d[1] =1.81 (Steketee et al., 2010) and d=1.98 (Tolin et al., 2007a). Ayers et al. (2011) also found a 14% significant reduction post-treatment improvement on the UHSS (Saxena et al., 2007). Results were more mixed for outcome on the HRS (Tolin, Frost, et al., 2010). Steketee et al. (2010) found a significant improvement for the whole sample after treatment (large ES; d=2.29). However, the waitlist control group also improved significantly at the 12 week comparison, though the CBT group improved significantly more. Turner et al. (2010) did not find a significant effect of treatment on mean HRS score, although they did have a small sample size (n=11) and used only the first 3 items of the HRS which limits comparability. Three studies looked at outcomes on the CIR (Frost et al., 2008). Significant mean improvements in clutter were reported by Tolin et al. (2007; large ES, d=2.85, 31% reduction) and Turner et al. (2010; mean 33.5% reduction), but not Ayers et al. (2011).

Three studies reported levels of clinically significant change (CSC[2]; Jacobson & Truax, 1991). Between 25-60% of participants were considered to have CSC in hoarding symptoms on the SI-R or HRS (Ayers et al., 2011; Steketee et al., 2010 ; Tolin et al., 2007a).

Of the two studies that included a follow-up measurement point, gains were maintained at 1 year follow up in one study (Muroff et al., 2014; Steketee et al., 2010) and the other found that some individual participants had 'relapsed'[3] by 6 months post-treatment, but overall mean SI-R scores remained significantly improved from baseline (Ayers et al., 2011).

Group CBT

Three of the group CBT studies assessed hoarding using questionnaire cut-offs and psychiatric interview. These studies found significant reductions on the SI-R (Frost et

[1]d =Cohen's d (Cohen, 1988). Cohen (1988) suggests interpreting effect sizes of 0.2 as small, 0.5 as medium and 0.8 and above as large.

[2] CSC defined in all studies using either Jacobson and Truax's (1991) criteria, or >30% reduction on at least one hoarding measure plus clinician rated improvement.

[3] Ayers et al. (2011) defined relapse as return to baseline scores

al., 2004), which ranged from 14% (Muroff et al., 2009) to 29.9% (compared to 9% in the control condition; Muroff et al., 2012). Overall ES, where reported, were large (d=1.31 (Gilliam et al., 2011), d=2.0 (Muroff et al., 2012), d=1.57 (Muroff et al., 2009)). Muroff et al. (2012) also found a significant improvement on the HRS (Tolin, Frost, et al., 2010) in the group CBT conditions (25.4-26.5%) compared to bibliotherapy control (8.6%). In contrast, Tolin et al. (2012), the only group CBT study to use proposed DSM-5 criteria, did not find any change on either the SI-R or the HRS. However, it should be noted that this study had a small sample size (n=6), which increases the chance of a type II error. The authors reported ES of d=0.48 on the SI-R and d=0.82 on the HRS-I, suggesting there were some modest improvements. Muroff et al. (2009) also found a significant improvement on the CIR after treatment (14% reduction). No significant changes were found for the SCI.

Two studies reported CSC. Gilliam et al. (2011) found 31% of completers and 23% of the intention-to-treat sample met criteria for CSC on the SI-R. Muroff et al. (2012) found 21.4% of group CBT without home assistance and 36.4% of group CBT plus home assistance participants met CSC criteria.

CBT-based bibliotherapy support groups

All three bibliotherapy group intervention studies defined hoarding according to questionnaire cut-offs. All reported significant reductions in hoarding symptoms as measured by the SI-R or HRS (improvement range 23-38%) with large ES (e.g. d=1.84, Frost et al., 2012). Frost et al. (2011, study 1 and 2) found that improvements were maintained at 1-month follow-up. Frost et al.'s (2012) study found that for the wait-list control group, HD symptoms increased over time. All studies also included the CIR and all found that levels of clutter significantly reduced from pre-post treatment (Frost et al., 2012, large ES d=1.84-1.91, 16-19% reduction (clinician and self-report); Frost et al., 2011, large ES d=1.88-2.85, pre-post 22.3-26% reduction). One study (Frost et al., 2011, Study 1) also included a 1-month follow-up but failed to identify any further improvements. Hoarding related cognitions measured by the SCI improved post-intervention for all studies (Frost et al., 2011 studies 1 & 2; Frost et al., 2012).

Frost et al. (2011, study 1) found 47% met criteria for CSC at post treatment, which increased to 59% at follow-up. Study 2 found 36% met criteria for CSC at post-treatment, which again increased at follow-up to 54%. In Frost et al. (2012), 30% met CSC criteria at post treatment.

3.7.2 Cognitive Remediation

Both studies using cognitive remediation used DSM-5 criteria to identify participants with HD. Findings were mixed with regard to the impact of cognitive remediation on HD symptoms. The two studies reported quite different aims and treatments. Ayers et al.'s (2014) study included exposure, which focused specifically on reducing hoarding severity whereas DiMauro et al.'s (2014) study did not include any elements of CBT. The heterogeneity in interventions precludes direct comparison between findings. Ayers and colleagues' (2014) study found significant reductions in hoarding symptoms measured by the SI-R (8.36% reduction, d=1.02, large ES), the UHSS (40.86% reduction, d=1.51, large ES), and the CIR (25.96% reduction, d=0.41, medium ES). DiMauro et al. (2014) did not find any significant effects on symptoms as measured by the SI-R.

3.7.3 Pharmacotherapy

Saxena et al. (2007) used the Y-BOCS (Goodman, 1989) as the main outcome measure, which is not hoarding specific, and the Frost and Hartl (1996) criteria for compulsive hoarding. They did, however, administer the UHSS to a subset of 19 hoarding participants. The UHSS was designed by the research team during the last 4 years of the study, hence was only administered to participants who took part in the latter part of the study. These 19 participants showed significant improvements post-treatment on the UHSS (24% reduction). Saxena et al. (2014), who used full DSM-5 criteria to diagnose HD, also found significant improvements on mean UHSS score (36% reduction, d=1.98, large ES) and on the SI-R (32% reduction, d=1.68, large ES). The non-hoarding OCD comparison group in Saxena et al. (2007) also showed significant improvements on the UHSS, which represented a greater (46%) reduction in scores than the hoarding group, indicating that the Paroxetine had a weaker effect for hoarding participants.

3.7.4 Other

Relatives intervention

Chasson et al. (2014) did not find a significant difference in relative-rated HRS scores at mid or post intervention. Mean scores decreased slightly with a medium ES (d=.37).

Online support group

Muroff et al.'s (2010) evaluation of an online support group had a substantial amount of missing data, making results difficult to interpret. In addition, participants 'self-identified' as meeting Frost and Hartl's (1996) criteria for compulsive hoarding, which was not verified by the researchers. The authors reported on a subgroup of participants who completed measures at all time points (all of whom were receiving the intervention), and also on any participants who remained in the study and provided data over 6 months (wait-list, long term members, and short term members of the website).

The participants with complete data showed significant improvements on the SI-R (n=23, 15.7% reduction) and the CIR (n=22, 18.2% reduction). Over a 6 month period, recent group members (members of the website for less than 3 months) showed significant reductions with medium ES's on the SI-R (d=.52) and the CIR (d=.47). Long-term members (over 3 months prior to study start) showed significant improvement on the CIR (small ES; d=.26), but not on the SI-R. The authors also found that the wait-listed group showed a significant improvement over 6 months on the SI-R. Over the total 15 months of the study, both long-term and recent members improved significantly on the SI-R, and recent members also improved significantly on the CIR.

3.8 Secondary outcomes by intervention type

As can be seen in table 3 (appendix 6), not all studies reported outcomes for anxiety, depression or functional impairment.

3.8.1 *CBT interventions*

Individual CBT

Two studies looked at the impact of individual CBT on emotional dysfunction. Ayers et al. (2011) found a significant decrease in depression symptoms after the intervention, which was maintained at 6 month follow-up. No significant effects were observed for anxiety. In contrast, Steketee et al. (2010) found no effect on either anxiety or depression using the same measures (BDI/BAI; Beck et al., 1996; Steer & Beck, 1997).

Two studies recorded levels of functional impairment, one using the ADL-H (Frost et al., 2013) and the other the SDS (Sheehan, 1983). Turner et al. (2010) found a significant reduction in difficulty on the ADL-H (24% reduction), whereas Ayers et al. (2011) found no significant difference on the SDS.

Group CBT

Three studies included outcomes on anxiety or depression and two on functional impairment using the ADL-H or SDS. Gilliam et al. (2011) found significant post-intervention improvement on anxiety and depression using the DASS (Lovibond & Lovibond, 1993). Muroff et al. (2009) found significant improvements in depression on the BDI, which was replicated compared to a bibliotherapy control condition in Muroff et al. (2012). Gilliam et al. (2011) found significant improvements at post intervention for both the SDS and ADL-H, and Muroff et al. (2012) also found improvements on the ADL-H after group CBT compared to bibliotherapy.

CBT-based bibliotherapy support groups

Both of Frost et al.'s (2011) studies included measures of depression, anxiety, and stress using the DASS. Neither study found a significant difference on any scale after the intervention or at follow-up. All studies reported outcomes on the ADL-H. Frost et al. (2011, study 1) found a significant decrease from pre-post, whereas study 2 found a significant decrease both at pre-post and at follow-up. Frost et al. (2012) found that ADL-H scores decreased significantly more for the treatment condition compared to the waitlist arm (30% improvement vs 2%). Their overall pre-post sample scores decreased significantly on the ADL-H.

3.8.2 Pharmacotherapy

Both pharmacotherapy trials found significant improvements on measures of depression and anxiety. Saxena et al. (2014) found substantial decrease in scores; 48% decrease in the mean Hamilton Depression Rating Scale (Hamilton, 1960b) score and a 43% decrease in the mean Hamilton Anxiety Scale (Hamilton, 1960a) score. Both studies also found significant decreases in OCD symptoms on the Y-BOCS (31-39% reduction), which was comparable to the decrease for the OCD group in Saxena et al. (2014).

3.8.3 Other

Relatives intervention

Chasson et al. (2014) found a significant improvement in the Family Response to Hoarding Scale (Steketee & Frost, 2013a) and the Family Member Impact Scale (Orford et al., 2005) at mid-treatment but not post. There was no change on the quality of life scale (Burckhardt et al., 1989).

4. Discussion

HD is a highly prevalent severe and enduring mental health condition associated with substantial distress and impairment for the person with HD and their families (APA, 2013). The current review aimed to explore and assess the quality of the available evidence regarding treatments for the symptoms of HD and related problems, in order to aid clinicians in identifying appropriate treatment for individuals with HD and to ascertain areas for future research. Seventeen studies were reviewed, comprising a range of treatment approaches, namely CBT (11 studies), pharmacology (2 studies), cognitive rehabilitation (2 studies), online support (1 study), and an intervention for family members (1 study). From the reviewed evidence it would seem fair to conclude there is no evidently superior treatment for HD.

Although thirteen studies reported statistically significant reductions in hoarding symptoms, with mostly large effect sizes on the main HD measures (range: d=0.52-2.29) and clutter (range: d=0.26-2.85), these reductions were modest (14-40%) and the majority of participants remained in the clinical range at the end of treatment. This difference between statistically and clinically meaningful change is important to consider, as although symptoms might improve somewhat, patients may still have

significant hoarding symptoms and levels of clutter at the end of treatment. Future research might usefully focus on patient reported outcome measures and functional outcomes to assess whether interventions result in meaningful change for the individual with HD. As other reviews have deduced, there is much room for development of alternative treatment modalities in HD and improving the acceptability and efficacy of current treatments (Muroff et al., 2011; Tolin et al., 2015).

The majority (11/17) of reviewed studies were CBT interventions, and all but two (Tolin et al., 2012; Turner et al., 2010) showed significant improvements in hoarding symptoms. Similarly to Tolin et al.'s (2015) meta-analysis, this review found comparable improvements between group and individual based CBT, and group bibliotherapy. However, bibliotherapy trials tended to use less stringent assessment of hoarding so it is possible these participants had less severe symptoms. Schmalisch et al. (2010) discuss the advantages of group based treatment for HD. They propose that the group setting may have benefits in reducing stigma and shame around hoarding, promoting motivation to change and reducing social isolation. Group treatment also has the advantage of being more cost efficient. A novel finding in this review, which adds to Tolin et al.'s (2015) findings, was that treatments other than CBT also improved HD symptoms by up to a third, such as Venlafaxine treatment (Saxena & Sumner, 2014) and a combined cognitive rehabilitation and CBT treatment (Ayers et al., 2014). There was some evidence that an online support group format could be helpful in improving hoarding symptoms, although this requires further study. More mixed results were found regarding improvement in clutter (as measured by the CIR). The Ayers et al. (2011) study, which used proposed DSM-5 criteria and thus may have had more severe cases, did not find a significant reduction after CBT. Tolin et al. (2015) suggested that clutter levels may take longer to reduce than other HD symptoms.

The four studies which failed to find a significant improvement in HD symptoms following intervention were characterised either by small sample sizes and large dropout rates (Tolin et al., 2012; Turner et al., 2010), or a lack of focus of the treatment on HD symptoms (Chasson et al., 2014; Dimauro et al., 2014). Dimauro

and colleague's (2014) primary aim was to improve associated neurocognitive deficits (i.e., attention, which did improve post-intervention) and their intervention did not include elements targeting HD symptoms. Their study did successfully improve participant attention post-intervention. Chasson and colleagues (2014) aimed to improve the experience of relatives in terms of impact of hoarding and family accommodation, and did not involve the individual with HD in the treatment.

As a secondary aim, this review sought to investigate the effects of interventions on comorbid psychiatric symptoms and functional impairment in HD. Once again, results were mixed, thus limiting the ability to draw firm conclusions about which treatments are most efficacious. Six of nine studies which measured levels of anxiety and depression in individuals with HD found an improvement. Both pharmacological studies showed a significant reduction in anxiety and depression, which is not particularly surprising given the known efficacy of SSRIs and SNRIs for these disorders (e.g. Zohar & Westenberg, 2000). The three group CBT studies and one individual CBT study also all found a positive improvement in comorbid symptoms. Across the four studies, the mechanisms of change remain unclear. However, it is possible that the element of peer-support and normalising could have contributed to improved outcomes in depression and anxiety after group CBT (Schmalisch et al., 2010; Whitfield, 2003; Yalom & Leszcz, 2005). On the other hand, the bibliotherapy-based support groups (Frost et al., 2011) did not show an improvement in anxiety or depression. This is an interesting difference, given that the manuals used for group CBT and CBT-based bibliotherapy (Tolin et al., 2007b; Steketee & Frost, 2007) follow similar formats. Neither includes a specific section on depression, although it is possible CBT skills learnt could have been generalised to other areas. It may be that the flexibility afforded by professionally facilitated groups played a role in improving depression.

Of seven papers which measured functioning or quality of life, all but one found an improvement. This finding might be expected as hoarding severity/clutter reduced, as items on the ADL-H refer to use of rooms in the home, living conditions, and safety. Interestingly in one study (Ayers et al., 2011), there were no reported functional improvements even though HD symptoms had improved.

An important finding of this review was the identification of a number of key issues in the current evidence base. Firstly, all studies were conducted in the USA by the same few research groups (e.g. Gail Steketee and Jordana Muroff at Boston University). This highlights the paucity of research into HD treatment in other countries and may impact on the generalisability of findings. Secondly, the 'diagnosis' of HD was not consistent across studies, which impacted on study comparisons. Many studies were conducted before the publication of the DSM-5, and thus may have included participants who would not have met full HD criteria, had less severe hoarding symptoms, or had hoarding symptoms in the context of other disorders. Thirdly, participants were mainly female and Caucasian, despite HD being at least as prevalent in men and hoarding symptoms being found in a range of cultures and ethnicities (Mataix-Cols et al., 2010). Fourthly, many of the studies did not include longer term follow-up to assess whether effects of treatments were enduring.

The quality assessment revealed that most studies were of low methodological quality, with all falling below the cut-off for adequate quality suggested by Wykes et al. (2008). Almost all of the studies under review had major methodological shortcomings, most notably small sample sizes and lack of control conditions. As has been noted in previous reviews (e.g. Muroff et al., 2011), attrition was relatively high and in some studies even close to 50% (Turner et al., 2010; Chasson et al., 2014). In addition, most studies recruited participants through public media advertisements (e.g. newspaper and TV adverts) which are likely to recruit a selective group of participants. It could be argued that such participants could have better insight into their problems and are possibly more motivated or amenable to change, particularly given what is known about low rates of treatment seeking in HD (Frost et al., 2010). Most studies were uncontrolled trials, which are prone to multiple sources of bias over and above RCTs (Reeves, Deeks, Higgins, & Wells, 2008). In uncontrolled trials, effects cannot be confidently attributed to the treatment, rather than nonspecific effects, time or regression to the mean. Most of the studies reviewed did not utilise methods to reduce other risks of bias, such as use of blind, independent assessors. Further, some researchers have warned that effect sizes can be inflated in small scale studies (Sterne, Gavaghan, & Egger, 2000) and that positive outcomes are likely

to be overestimated in non-randomised trials (Schulz, Chalmers, Hayes, & Altman, 1995). A major priority for future research in this area will be to produce better quality trials, taking account of the shortcomings identified in this review.

Of the studies that were RCTs, most used waitlist controls which, whilst being pragmatic, are not necessarily the ideal comparator for psychological trials. Waitlist controls do not control for non-specific effects of treatments and those being randomised to waitlist may experience unintended effects, such as symptoms becoming worse and therefore overestimating the effect of the treatment condition, or discouraging participants from staying in the trial (e.g. Basham, 1986; Hart, Fann, & Novack, 2008; Mohr et al., 2009). The importance of an active control group is further emphasised by findings by Steketee et al. (2010) that their wait-list control group also improved significantly by week 12 of the study. Studies which used active controls (Muroff et al., 2012; Dimauro et al., 2014) provide more convincing evidence of treatment efficacy.

Another source of heterogeneity across the studies was the use of different outcome measures for HD symptoms. Thus far there is no 'gold standard' measure for HD, although the most widely used in research are the SI-R (Frost et al., 2004), HRS (Tolin, Frost, et al., 2010), and the UHSS (Saxena et al., 2007). A recent study (Saxena, Ayers, Dozier, & Maidment, 2015) compared the UHSS and the SI-R and found that they did not fully correlate, indicating that they may be measuring different aspects of HD. Additionally, the UHSS contains some items non-specific to HD, such as perfectionism, procrastination, and indecisiveness. In one of the reviewed studies (Saxena et al., 2007), the 'non-hoarding OCD' comparison group actually experienced a greater reduction in scores on the UHSS than the hoarding group. This could be interpreted in a number of ways, including the non-specificity of some items on the questionnaire, the greater treatment resistance in the HD group or simply a chance finding, since only six non-hoarding patients completed the UHSS. Nevertheless, it may be useful for future research to investigate the specificity of the questionnaires measuring hoarding symptoms.

4.1 Strengths and Limitations

The key strengths of the current review include the systematic approach to the search methods, designed to ensure a fair and thorough representation of the current literature. All included studies were discussed between two researchers and checked against inclusion criteria. Another strength was the use of an established quality assessment tool to assess the methodological strengths and weaknesses of the studies in the areas. The quality assessment revealed many flaws in the research and improvements needed for future studies. The shortcomings identified also suggest the appropriateness of a systematic review over the meta-analytic approach adopted by Tolin et al. (2015); the results of a meta-analysis could be misleading due to significant heterogeneity in study design and quality (Ahlbom, 1993; Bailar, 1995). Another strength in using systematic review methodology allowed the comparison of studies trialling different treatment approaches, which revealed similar efficacy rates between various treatments.

The review also had some limitations. First, we included studies which did not use the full DSM-5 diagnostic criteria to identify HD cases, meaning that these studies may include participants who did not have HD and indeed whose hoarding symptoms may have been less severe, or better explained by other mental health problems. There are still very few research studies using the diagnostic criteria, as the DSM-5 was only published two years ago. Second, studies with a negative or non-significant outcome have a tendency to not be published (the "file drawer effect"), and thus were not included in the review, which may have led to a bias in the conclusions. Third, the inclusion of both RCTs and uncontrolled trials was a limitation. The Cochrane collaboration (2011) advise against including both in the same review, due to the heterogeneity of study designs and outcomes, which make a summary of the data difficult to interpret. However, in this case the aim was to be as comprehensive as possible in study selection, and given the scarce research in the field and HD having just been accepted as a disorder in its own right, very few RCTs existed. The fourth limitation was related to the quality assessment measure. Many of the questions on the CTAM are related to an RCT design (e.g. randomisation, blinding) and therefore the uncontrolled or non-randomised trials consistently lost

points on these items. This may mean that the CTAM results are not directly comparable to other published systematic reviews and meta-analyses which have used this tool. Other reviews which have used the CTAM have generally reviewed RCTs only (e.g. Lobban et al., 2013; van der Gaag et al., 2013; Wykes, Steel, Everitt, & Tarrier, 2008) and therefore can be expected to have higher scores. However, it is important to note that the CTAM was not designed specifically for RCTs only (Tarrier & Wykes, 2004) and the low scores on the CTAM in this review do reflect the poorer quality of studies in this field so far. Higher quality RCTs are required to extend the evidence base.

4.2 Clinical Implications

The present review offers a number of implications for clinical practice. It has shown that treatments for HD, although still in the early stages of development and despite the generally low methodological quality of the reviewed studies, can be effective in reducing the symptoms and impact of HD on patients' lives. The most reliable evidence thus far is for group or individual CBT following a HD-specific protocol (e.g. Muroff, Underwood, & Steketee, 2014; Steketee & Frost, 2013). Further evidence comes from single-case experimental designs (e.g. Pollock et al., 2014) which provide ecologically valid and scientifically rigorous methodology for testing interventions and indicate that individual CBT for HD can be effective in clinical settings.

This review also indicates that interventions other than CBT could be useful clinically. Cognitive remediation, for instance, may be a useful adjunct to CBT in improving the cognitive features of HD such as attention. In a qualitative study of patient and therapist perspectives on CBT for HD, therapists identified poor attention and executive functioning as a cause of poorer outcome in CBT (Ayers, Bratiotis, Saxena, & Wetherell, 2012). Many studies have shown cognitive remediation to be helpful in other disorders, such as schizophrenia, particularly as an addition to other psychological interventions where cognitive deficits are observed (e.g. McGurk, Twamley, Sitzer, McHugo, & Mueser, 2007; Medalia & Saperstein, 2013; Wykes, Huddy, Cellard, McGurk, & Czobor, 2011). The results of this review indicate that SSRIs (namely Venlafaxine, which had better acceptability, Saxena & Sumner, 2014) could also be a useful addition to treatment in reducing HD symptoms and

associated anxiety and depression. Reductions in depression and anxiety could potentially enhance motivation for engaging with treatment for hoarding behaviours, and multimodal interventions have shown promising outcomes for people with OCD and hoarding behaviours (Saxena & Maidment, 2004). Further research is required to determine whether combined treatments are more effective than either medication or psychological treatment alone.

The intervention for relatives showed promise in reducing the impact of HD on the family (Chasson et al., 2014), but requires replication and further study to determine longer term effects and mechanisms for positive outcomes. Interventions involving both the individual with HD and family members are yet to be studied, which have the potential to improve outcomes in terms of HD symptoms. Increasingly mental health services are recommended to offer support for relatives of service users, which can be helpful in reducing family distress and burden in severe mental health conditions (e.g. Gaebel, Weinmann, Sartorius, Rutz, & McIntyre, 2005; Yesufu-Udechuku et al., 2015). Recognition of the needs of families of people with HD will be important in developing clinical services. As well as engaging families, it is likely that treatment services will need to be inter-agency and multidisciplinary to address the various challenges and risks of working with HD (Bratiotis, 2013). As discussed, the societal costs of HD are significant and thus investment in a broader range of interventions will be cost effective in the longer term.

A major challenge for developing services is likely to be engaging with people with HD, as many are not help seeking and/or aware of the severity of their needs or impact on others (e.g. Koenig, Spano, Leiste, Holmes, & Macmillan, 2014). Advice and guidance from people with lived experience of HD would be valuable in attempting to address some of these barriers. Another challenge for services will be the cost of providing the current manualised CBT, which consists of a larger number of sessions than is typical for other disorders and includes home visits. Given that specialist hoarding services are very few, this may be challenging for people with HD who do want psychological therapy. This reflects similar issues observed in other severe mental conditions such as schizophrenia, where the allocated resources and numbers of skilled therapists available are unable to meet the needs and demands

for therapy access (The Schizophrenia Commission, 2012). Consequently, equitable access is patchy and differs from one geographical region to another (Haddock et al., 2014).

Shorter group based CBT may be an alternative option. Home visits can also be difficult to provide if clients live far from the treatment centre, which is leading researchers to look into creative means of delivering CBT for HD using digital based methods, such as via webcam (Muroff, Steketee, & Frost, 2009). In addition, there are few clinicians trained in CBT for HD currently, and thus alternative treatment modalities, such as self-help via bibliotherapy, Internet-delivered CBT, or Internet support forums are likely to be the most accessible form of treatment. Some authors (e.g. Muroff, 2011) have suggested a 'stepped care' model for treatment of HD, with lower intensity treatments such as self-help for less severe cases, building up to individual treatment delivered by clinicians.

4.3 Directions for future research

This review has highlighted a number of areas which future research should address. In terms of methodology, better quality trials including RCTs are recommended to further the evidence base for CBT and other emerging treatments. RCTs should ideally use blinding for randomisation and assessment, employ assessors independent of the treatment team to reduce risk of bias and assess therapist competence and fidelity to treatment protocols. Studies should be sufficiently powered to detect treatment effects and use appropriate active control conditions, such as the self-help treatment condition used by Muroff et al. (2012). Given than HD is mainly a long-term condition, follow-up assessment points will be crucial to assess whether effects of treatment endure beyond the study period. Assessment of clutter after treatment and at follow-up will also be important, particularly as clutter does not always reduce substantially by the end of treatment and is a more 'objective' measure of symptom reduction than self-report (Frost et al., 2008). Single case experimental designs also make a contribution to the HD treatment literature and a review of these is needed in the future.

A key area for future research to address will be improving the acceptability of treatments, to increase uptake of interventions and reduce dropout rates in trials. It

will be important to assess reasons for dropout and consult with service users on the best ways to aid engagement. Future research should aim to improve on and revise existing protocols, as well as drawing on the literature in other disorders with similar features to develop novel treatments. For example, Tompkins has recently developed a harm reduction approach (Tompkins, 2011, 2015), based on interventions in the addictions field where people may not wish to stop their problem behaviour altogether (as is often the case in HD due to reduced insight into their condition), but instead can work on ways of improving safety and limiting the negative effects of the behaviour.

Family interventions have proved helpful in other disorders with similar features, such as in OCD and psychosis (e.g. Onwumere, Bebbington, & Kuipers, 2011; Pharoah, Mari, Rathbone, & Wong, 2010; Steketee & Van Noppen, 2003). As well as the Chasson et al., (2014) study described here, Sampson and colleagues (Sampson et al., 2012) conducted a qualitative study which demonstrated the usefulness of a brief group based carer intervention for relatives of people who hoard; relatives reported better understanding of HD and improved interactions with their hoarding relative after the intervention. Such interventions merit further study, given the substantial impact of HD on family members and the potential of family members to motivate people with HD to seek treatment (Chasson et al., 2014; Tompkins, 2011). In addition, improving relationships between family members would reduce the social isolation often seen in people with HD (Medard & Kellett, 2014).

Additionally, combined pharmacotherapy and psychological therapy have the potential to improve treatment gains. Saxena and colleagues (Saxena et al., 2002) reported that people with OCD and hoarding symptoms showed significant improvements after intensive CBT plus medication and psychosocial rehabilitation. It remains to be seen whether people with HD would respond to multimodal or more intensive treatments. Furthermore, as individual and group CBT both appear to be helpful to a certain extent, it would be prudent to directly compare the two.

As this review has identified that a range of different treatments could be effective in treating HD, an important future step would be to determine what the active components are of different treatments. Further research should be conducted to

determine what predicts which patients will respond best to treatment, and which treatments are appropriate for whom. Tolin and colleagues (2015) looked at some potential moderators of response to CBT, but other plausible moderators or mediators are worthy of further investigation such as cognitive functioning and baseline level of insight. In other disorders it has been identified that people with supportive relationships respond better to psychological therapies independent of the treatment modality (Driessen & Hollon, 2010; Garety et al., 2008). In OCD, family dysfunction, negative interactions, and family accommodation predict worse treatment outcomes (Keeley, Storch, Merlo, & Geffken, 2008). Future research could usefully investigate the role of relatives in the course and treatment of HD, as well as the effect of carer only interventions in improving understanding and interactions in families.

Finally, this review has revealed a lack of consistency in the diagnosis and measurement methods used in the HD research field. Now that the DSM-5 criteria exists, all future studies should strictly diagnose HD, including ruling out other possible causes of hoarding symptoms, using standardised interviews such as the Structured Interview for Hoarding Disorder (SIHD; Nordsletten et al., 2013). Researchers should also aim to reach a consensus on which instruments have best specificity, validity, and reliability to measure the response of HD to treatment.

4.4 Conclusion

HD is a newly defined mental health condition for which new specialised treatments are developing. This review was the first study to comprehensively qualitatively review both psychological and pharmacological treatments for hoarding, including family treatments, since the publication of the DSM-5 in 2013. The findings suggest promising outcomes for the efficacy of a range of interventions for HD. However, the current treatments are not effective enough, since most patients remain in the clinical range at the end of treatment. Given what we know about the significant impact of HD on the individual and their families, the associated health and safety risks and the consequences for public services, it is vital that research into treatments continues to develop and work toward better efficacy.

5. Bibliography

Ahlbom, A. (1993). Pooling Epidemiologic Studies. *Epidemiology*, *4*, 283–284.

American Psychiatric Association. (2000). *Diagnostic and statistical manual of mental disorders (4th ed.)*. Washington, D.C.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders: DSM-5.* (5th ed.). Washington, D.C.: American Psychiatric Association.

Ayers, C. R., Bratiotis, C., Saxena, S., & Wetherell, J. L. (2012). Therapist and patient perspectives on cognitive-behavioral therapy for older adults with hoarding disorder: A collective case study. *Aging & Mental Health*, *16*(7), 915–921. doi:10.1080/13607863.2012.678480

Ayers, C. R., Loebach, J., Golshan, S., & Saxena, S. (2011). Cognitive-behavioral therapy for geriatric compulsive hoarding. *Behaviour Research and Therapy*, *49*(10), 689–694. doi:10.1016/j.brat.2011.07.002

Ayers, C. R., Saxena, S., Espejo, E., Twamley, E. W., Granholm, E., & Wetherell, J. L. (2014). Novel Treatment for Geriatric Hoarding Disorder: An Open Trial of Cognitive Rehabilitation Paired with Behaviour Therapy. *American Journal of Psychiatry*, *22*(3), 248–252. doi:10.1016/j.jagp.2013.02.010.Novel

Ayers, C. R., Saxena, S., Golshan, S., & Wetherell, J. L. (2010). Age at onset and clinical features of late life compulsive hoarding. *International Journal of Geriatric Psychiatry*, *25*(2), 142–9. doi:10.1002/gps.2310

Bailar, J. C. (1995). The practice of meta-analysis. *Journal of Clinical Epidemiology*, *48*(1), 149. doi:10.1016/0895-4356(94)00149-K

Basham, R. B. (1986). Scientific and practical advantages of comparative design in psychotherapy outcome research. *Journal of Consulting and Clinical Psychology*, *54*(1), 89–94. doi:10.1037/0022-006X.54.1.88

Bech, P. (1981). Rating scales for affective disorders: their validity and consistency. *Acta Psychiatrica Scandinavica. Supplementum*, *295*, 1–101. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/7044046

Beck, A. T., Steer, R. A., & Brown, G. K. (1996). *Manual for the Beck Depression Inventory-II.* San Antonio, TX: Psychological Corporation.

Beck, A. T., Ward, C. H., Mendelson, M., Mock, J., & Erbaugh, J. (1961). An inventory for measuring depression. *Archives of General Psychiatry*, *4*, 561–571.

Beneke, M., & Rasmus, W. (1992). “Clinical Global Impressions” (ECDEU): some critical comments. *Pharmacopsychiatry*, *25*(4), 171–6. doi:10.1055/s-2007-1014401

Bloch, M. H., Bartley, C. a, Zipperer, L., Jakubovski, E., Landeros-Weisenberger, a, Pittenger, C., & Leckman, J. F. (2014). Meta-analysis: hoarding symptoms associated with poor treatment outcome in obsessive-compulsive disorder. *Molecular Psychiatry*, *19*(9), 1025–1030. doi:10.1038/mp.2014.50

Bratiotis, C. (2013). Community hoarding task forces: A comparative case study of five task forces in the United States. *Health and Social Care in the Community*, *21*(3), 245–253. doi:10.1111/hsc.12010

Bubrick, J., Neziroglu, F., & Yaryura-Tobias, J. (2004). *Overcoming Compulsive Hoarding: Why You Save and How You Can Stop*. Oakland, CA: New Harbinger Publications. Retrieved from http://books.google.com/books?hl=en&lr=&id=aPJ4gmKSY4QC&pgis=1

Burckhardt, C. S., Woods, S. L., Schultz, A. A., & Ziebarth, D. M. (1989). Quality of life of adults with chronic illness: a psychometric study. *Research in Nursing & Health*, *12*(6), 347–54. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/2602575

Chasson, G. S., Carpenter, A., Ewing, J., Gibby, B., & Lee, N. (2014). Empowering families to help a loved one with Hoarding Disorder: Pilot study of Family-As-Motivators training. *Behaviour Research and Therapy*, *63*, 9–16. doi:10.1016/j.brat.2014.08.016

Chasson, G. S., Ewing, J., Gibby, B., & Carpenter, A. (2013). *Family-As-Motivators training implementaion manual*. Unpublished Manual.

Cohen, J. (1988). *Statistical Power Analysis for the Behavioral Sciences* (Second.). Lawrence Erlbaum Associates.

Cook, D. J. (1997). Systematic Reviews: Synthesis of Best Evidence for Clinical Decisions. *Annals of Internal Medicine*, *126*(5), 376. doi:10.7326/0003-4819-126-5-199703010-00006

Cromer, K. R., Schmidt, N. B., & Murphy, D. L. (2007). Do traumatic events influence the clinical expression of compulsive hoarding? *Behaviour Research and Therapy*, *45*, 2581–2592. doi:10.1016/j.brat.2007.06.005

Dahlke, F., Lohaus, A., & Gutzmann, H. (1992). Reliability and clinical concepts underlying global judgments in dementia: implications for clinical research. *Psychopharmacology Bulletin*, *28*(4), 425–32. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/1296220

DeCoster, J. (2012). *Spreadsheet for converting effect size measures.* Retrieved May 4, 2015, from http://www.stat-help.com/spreadsheets/Converting effect sizes 2012-06-19.xls

Dimauro, J., Genova, M., Tolin, D. F., & Kurtz, M. M. (2014). Cognitive remediation for neuropsychological impairment in hoarding disorder : A pilot study. *Journal of Obsessive-Compulsive and Related Disorders*, *3*(2), 132–138. doi:10.1016/j.jocrd.2014.03.006

Driessen, E., & Hollon, S. D. (2010). Cognitive behavioral therapy for mood disorders: efficacy, moderators and mediators. *The Psychiatric Clinics of North America*, *33*(3), 537–55. doi:10.1016/j.psc.2010.04.005

Drury, H., Ajmi, S., Fernández de la Cruz, L., Nordsletten, A. E., & Mataix-Cols, D. (2014). Caregiver burden, family accommodation, health, and well-being in relatives of individuals with hoarding disorder. *Journal of Affective Disorders*, *159*, 7–14. doi:10.1016/j.jad.2014.01.023

Falloon, I. R. H. (2003). Family interventions for mental disorders: efficacy and effectiveness. *World Psychiatry : Official Journal of the World Psychiatric Association (WPA)*, *2*(1), 20–8. Retrieved from http://www.pubmedcentral.nih.gov/articlerender.fcgi?artid=1525058&tool=pmcentrez&rendertype=abstract

Forkmann, T., Scherer, A., Boecker, M., Pawelzik, M., Jostes, R., & Gauggel, S. (2011). The Clinical Global Impression Scale and the influence of patient or staff perspective on outcome. *BMC Psychiatry*, *11*, 83. doi:10.1186/1471-244X-11-83

Frost, R. O. (2010). Treatment of Hoarding. *Expert Review of Neurotheraputics*, *10*(2), 251–261.

Frost, R. O., & Gross, R. C. (1993). The hoarding of possessions. *Behaviour Research and Therapy*, *31*(4), 367–381. doi:10.1016/0005-7967(93)90094-B

Frost, R. O., & Hartl, T. L. (1996). A cognitive-behavioral model of compulsive hoarding. *Behaviour Research and Therapy*, *34*(4), 341–50. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/8871366

Frost, R. O., Hartl, T. L., Christian, R., & Williams, N. (1995). The value of possessions in compulsive hoarding: Patterns of use and attachment. *Behaviour Research and Therapy*, *33*(8), 897–902. doi:10.1016/0005-7967(95)00043-W

Frost, R. O., Hristova, V., Steketee, G., & Tolin, D. F. (2013). Activities of daily living scale in hoarding disorder. *Journal of Obsessive-Compulsive and Related Disorders*, *2*(2), 85–90. doi:10.1016/j.jocrd.2012.12.004

Frost, R. O., Patronek, G., & Rosenfield, E. (2011). Comparison of object and animal hoarding. *Depression and Anxiety*, *28*(10), 885–91. doi:10.1002/da.20826

Frost, R. O., Pekareva-Kochergina, A., & Maxner, S. (2011). The effectiveness of a biblio-based support group for hoarding disorder. *Behaviour Research and Therapy*, *49*(10), 628–634. doi:10.1016/j.brat.2011.06.010

Frost, R. O., Ruby, D., & Shuer, L. J. (2012). The Buried in Treasures Workshop: Waitlist Control Trial of Facilitated Support Groups for Hoarding. *Behaviour Research and Therapy*, *50*(11), 661–667. doi:10.1016/j.brat.2012.08.004.The

Frost, R. O., Steketee, G., & Grisham, J. (2004). Measurement of compulsive hoarding: saving inventory-revised. *Behaviour Research and Therapy*, *42*(10), 1163–82. doi:10.1016/j.brat.2003.07.006

Frost, R. O., Steketee, G., & Tolin, D. F. (2011). Comorbidity in hoarding disorder. *Depression and Anxiety*, *28*(10), 876–84. doi:10.1002/da.20861

Frost, R. O., Steketee, G., Tolin, D. F., & Renaud, S. (2008). Development and Validation of the Clutter Image Rating. *Journal of Psychopathology and Behavioral Assessment*, *30*(3), 193–203. doi:10.1007/s10862-007-9068-7

Frost, R. O., Steketee, G., & Williams, L. (2000). Hoarding: a community health problem. *Health & Social Care in the Community*, *8*(4), 229–234. doi:10.1046/j.1365-2524.2000.00245.x

Frost, R. O., Tolin, D. F., & Maltby, N. (2010). Insight-Related Challenges in the Treatment of Hoarding. *Cognitive and Behavioral Practice*, *17*(4), 404–413. doi:10.1016/j.cbpra.2009.07.004

Frost, R. O., Tolin, D. F., Steketee, G., Fitch, K. E., & Selbo-Bruns, A. B. (2009). Excessive Acquisition in Hoarding. *Journal of Anxiety Disorders*, *23*(5), 632–639. doi:10.1016/j.biotechadv.2011.08.021.Secreted

Frost, R. O., Tolin, D. F., Steketee, G., & Oh, M. (2011). Indecisiveness and Hoarding. *International Journal of Cognitive Therapy*, *4*(3), 253–262. doi:10.1521/ijct.2011.4.3.253

Gaebel, W., Weinmann, S., Sartorius, N., Rutz, W., & McIntyre, J. S. (2005). Schizophrenia practice guidelines: international survey and comparison. *The British Journal of Psychiatry : The Journal of Mental Science*, *187*(3), 248–55. doi:10.1192/bjp.187.3.248

Garety, P. A., Fowler, D. G., Freeman, D., Bebbington, P., Dunn, G., & Kuipers, E. (2008). Cognitive--behavioural therapy and family intervention for relapse prevention and symptom reduction in psychosis: randomised controlled trial. *The British Journal of Psychiatry : The Journal of Mental Science*, *192*(6), 412–23. doi:10.1192/bjp.bp.107.043570

Gilliam, C., Hannan, S. E., Norberg, M. M., Morrison, S., Villavicencio, A., & Dufresne, D. (2009). Group cognitive behavioral therapy for compulsive hoarding: An

effectiveness study. In *Paper presented at the Association of Behavioral and Cognitive Therapies*. New York, NY.

Gilliam, C. M., Norberg, M. M., Villavicencio, A., Morrison, S., Hannan, S. E., & Tolin, D. F. (2011). Group cognitive-behavioral therapy for hoarding disorder: An open trial. *Behaviour Research and Therapy*, *49*(11), 802–807. doi:10.1016/j.brat.2011.08.008

Goodman, W. K. (1989). The Yale-Brown Obsessive Compulsive Scale. *Archives of General Psychiatry*, *46*(11), 1006. doi:10.1001/archpsyc.1989.01810110048007

Grisham, J. R., Frost, R. O., Steketee, G., Kim, H.-J., & Hood, S. (2006). Age of onset of compulsive hoarding. *Journal of Anxiety Disorders*, *20*(5), 675–86. doi:10.1016/j.janxdis.2005.07.004

Grisham, J. R., Frost, R. O., Steketee, G., Kim, H.-J., Tarkoff, A., & Hood, S. (2009). Formation of attachment to possessions in compulsive hoarding. *Journal of Anxiety Disorders*, *23*(3), 357–61. doi:10.1016/j.janxdis.2008.12.006

Grisham, J. R., Norberg, M. M., Williams, A. D., Certoma, S. P., & Kadib, R. (2010). Categorization and cognitive deficits in compulsive hoarding. *Behaviour Research and Therapy*, *48*(9), 866–72. doi:10.1016/j.brat.2010.05.011

Guy, W. (Ed.). (1976). *Clinical Global Impression. ECDEU Assessment Manual for Psychopharmacology.* Rockville, MD: US Department of Heath, Education, and Welfare Public Health Service Alcohol, Drug Abuse, and Mental Health Administration.

Haddock, G., Eisner, E., Boone, C., Davies, G., Coogan, C., & Barrowclough, C. (2014). An investigation of the implementation of NICE-recommended CBT interventions for people with schizophrenia. *Journal of Mental Health*, *8237*, 1–4. doi:10.3109/09638237.2013.869571

Hall, B. J., Tolin, D. F., Frost, R. O., & Steketee, G. (2013). An exploration of comorbid symptoms and clinical correlates of clinically significant hoarding symptoms. *Depression and Anxiety*, *30*(1), 67–76. doi:10.1002/da.22015

Hamilton, M. (1960a). A rating scale for anxiety. *Journal of Neurology, Neurosurgery, and Psychiatry*, *23*, 56–62.

Hamilton, M. (1960b). Diagnosis and rating scale for depression. *British Journal of Psychiatry*, *3*, 76–79.

Hart, T., Fann, J. R., & Novack, T. A. (2008). The dilemma of the control condition in experience-based cognitive and behavioural treatment research. *Neuropsychological Rehabilitation*, *18*(1), 1–21. doi:10.1080/09602010601082359

Hartl, T. L., Duffany, S. R., Allen, G. J., Steketee, G., & Frost, R. O. (2005). Relationships among compulsive hoarding, trauma, and attention-deficit/ hyperactivity disorder. *Behaviour Research and Therapy*, *43*, 269–276. doi:10.1016/j.brat.2004.02.002

Hartl, T. L., & Frost, R. O. (1999). Cognitive-behavioral treatment of compulsive hoarding: A multiple baseline experimental case study. *Behaviour Research and Therapy*, *37*, 451–461. doi:10.1016/S0005-7967(98)00130-2

Hartl, T. L., Frost, R. O., Allen, G. J., Deckersbach, T., Steketee, G., Duffany, S. R., & Savage, C. R. (2004). Actual and perceived memory deficits in individuals with compulsive hoarding. *Depression and Anxiety*, *20*(2), 59–69. doi:10.1002/da.20010

Iervolino, A. C., Ph, D., Perroud, N., Fullana, M. A., Guipponi, M., Cherkas, L., … Mataix-cols, D. (2009). Prevalence and Heritability of Compulsive Hoarding : A Twin Study. *American Journal of Psychiatry*, *166*(October), 1156–1161.

Ivanov, V. Z., Mataix-Cols, D., Serlachius, E., Lichtenstein, P., Anckarsäter, H., Chang, Z., … Rück, C. (2013). Prevalence, Comorbidity and Heritability of Hoarding Symptoms in Adolescence: A Population Based Twin Study in 15-Year Olds. *PLoS ONE*, *8*(7), e69140. doi:10.1371/journal.pone.0069140

Jacobson, N. S., & Truax, P. (1991). Clinical significance: A statistical approach to defining meaningful change in psychotherapy research. *Journal of Consulting and Clinical Psychology*, *59*(1), 12–19.

Keeley, M. L., Storch, E. A., Merlo, L. J., & Geffken, G. R. (2008). Clinical predictors of response to cognitive-behavioral therapy for obsessive-compulsive disorder. *Clinical Psychology Review*, *28*(1), 118–30. doi:10.1016/j.cpr.2007.04.003

Kellett, S. (2006). The Treatment of Compulsive Hoarding with Object-Affect Fusion Informed CBT: Initial Experimental Case Evidence. *Behavioural and Cognitive Psychotherapy, 24, 481-485.* doi:10.1017/S1352465806003006

Kellett, S., Greenhalgh, R., Beail, N., & Ridgway, N. (2010). Compulsive hoarding: an interpretative phenomenological analysis. *Behavioural and Cognitive Psychotherapy*, *38*(2), 141–55. doi:10.1017/S1352465809990622

Koenig, T. L., Spano, R., Leiste, M. R., Holmes, R., & Macmillan, K. R. (2014). Social Work in Mental Health Multidisciplinary Teams' Practice Strategies With Older Adult Clients Who Hoard. *Social Work in Mental Health*, *12*(1), 81–97. doi:10.1080/15332985.2013.831390

Landau, D., Iervolino, A. C., Pertusa, A., Santo, S., Singh, S., & Mataix-Cols, D. (2011). Stressful life events and material deprivation in hoarding disorder. *Journal of Anxiety Disorders*, *25*(2), 192–202. doi:10.1016/j.janxdis.2010.09.002

Lobban, F., Postlethwaite, A., Glentworth, D., Pinfold, V., Wainwright, L., Dunn, G., … Haddock, G. (2013). A systematic review of randomised controlled trials of interventions reporting outcomes for relatives of people with psychosis. *Clinical Psychology Review*, *33*(3), 372–382. doi:10.1016/j.cpr.2012.12.004

Lovibond, S. H., & Lovibond, P. F. (1993). *Manual for the Depression Anxiety Stress Scales (DASS).* Psychology Foundation Monograph.

Mataix-Cols, D., Billotti, D., Fernández de la Cruz, L., & Nordsletten, A. E. (2013). The London field trial for hoarding disorder. *Psychological Medicine*, *43*(4), 837–47. doi:10.1017/S0033291712001560

Mataix-Cols, D., & Fernández de la Cruz, L. (2014). Diagnosis of Hoarding Disorder. In R. O. Frost & G. Steketee (Eds.), *Oxford Handbook of Hoarding and Acquiring* (1st ed., pp. 43–58). New York: Oxford University Press.

Mataix-Cols, D., Frost, R. O., Pertusa, A., Clark, L. A., Saxena, S., Leckman, J. F., … Wilhelm, S. (2010). Hoarding disorder: a new diagnosis for DSM-V? *Depression and Anxiety*, *27*(6), 556–72. doi:10.1002/da.20693

Mataix-Cols, D., Marks, I. M., Greist, J. H., Kobak, K. A., & Baer, L. (2002). Obsessive-compulsive symptom dimensions as predictors of compliance with and response to behaviour therapy: Results from a controlled trial. *Psychotherapy and Psychosomatics*, *71*(5), 255–262.

Mataix-Cols, D., Rauch, S. L., Manzo, P. A., Jenike, M. A., & Baer, L. (1999). Use of Factor-Analyzed Symptom Dimensions to Predict Outcome With Serotonin Reuptake Inhibitors and Placebo in the Treatment of Obsessive-Compulsive Disorder. *American Journal of Psychiatry*, *156*(9), 1409–1416. Retrieved from http://ajp.psychiatryonline.org/doi/abs/10.1176/ajp.156.9.1409

Mathews, C. A., Nievergelt, C. M., Azzam, A., Garrido, H., Chavira, D. A., Wessel, J., … Schork, N. J. (2007). Heritability and clinical features of multigenerational families with obsessive-compulsive disorder and hoarding. *American Journal of Medical Genetics. Part B, Neuropsychiatric Genetics : The Official Publication of the International Society of Psychiatric Genetics*, *144B*(2), 174–82. doi:10.1002/ajmg.b.30370

Matsunaga, H., Hayashida, K., Kiriike, N., Nagata, T., & Stein, D. J. (2010). Clinical Features and Treatment Characteristics of Compulsive Hoarding in Japanese Patients with Obsessive-Compulsive Disorder. *CNS Spectrums*, *15*(04), 258–266. doi:10.1017/S1092852900000092

McGurk, S. R., Twamley, E. W., Sitzer, D. I., McHugo, G. J., & Mueser, K. T. (2007). A Meta-Analysis of Cognitive Remediation in Schizophrenia. *Am J PsychiatryAm J Psychiatry*, *164*, 1791–1802.

Medalia, A., & Saperstein, A. M. (2013). Does cognitive remediation for schizophrenia improve functional outcomes? *Current Opinion in Psychiatry*, *26*(2), 151–7. doi:10.1097/YCO.0b013e32835dcbd4

Medard, E., & Kellett, S. (2014). The Role of Adult Attachment and Social Support in Hoarding Disorder. *Behavioural and Cognitive Psychotherapy*, *42*, 1–5. doi:10.1017/S1352465813000659

Miller, W. R., & Rollnick, S. (2002). *Motivational Interviewing, Second Edition: Preparing People for Change* (2nd ed.). New York: Guildford Press.

Mohr, D. C., Spring, B., Freedland, K. E., Beckner, V., Arean, P., Hollon, S. D., … Kaplan, R. (2009). The selection and design of control conditions for randomized controlled trials of psychological interventions. *Psychotherapy and Psychosomatics*, *78*, 275–284. doi:10.1159/000228248

Mueller, A., Mitchell, J. E., Crosby, R. D., Glaesmer, H., & de Zwaan, M. (2009). The prevalence of compulsive hoarding and its association with compulsive buying in a German population-based sample. *Behaviour Research and Therapy*, *47*(8), 705–709. doi:10.1016/j.brat.2009.04.005

Muroff, J. (2011). Thinking outside the box: Novel delivery methods for treating hoarding. *International Journal of Cognitive Therapy*, *4*(3), 280–296. doi:10.1521/ijct.2011.4.3.280

Muroff, J., Bratiotis, C., & Steketee, G. (2011). Treatment for Hoarding Behaviors: A Review of the Evidence. *Clinical Social Work Journal*, *39*(4), 406–423. doi:10.1007/s10615-010-0311-4

Muroff, J., Steketee, G., & Bratiotis, C. (2010). Enhancing group cognitive behavioral treatment for hoarding: A pilot study. In *Paper presented at the World Congress of Cognitive and Behavioral Therapies.* Boston, MA.

Muroff, J., Steketee, G., Bratiotis, C., & Ross, A. (2012). Group cognitive and behavioral therapy and bibliotherapy for hoarding: a pilot trial. *Depression and Anxiety*, *29*(7), 597–604. doi:10.1002/da.21923

Muroff, J., Steketee, G., & Frost, R. (2009). Cognitive Behavioral Treatment Delivered via Webcam. In *Paper presented at the Annual Meeting of the Association for Behavioral and Cognitive Therapies.* New York, NY.

Muroff, J., Steketee, G., Frost, R. O., & Tolin, D. F. (2014). Cognitive Behaviour Therapy For Hoarding Disorder : Follow-up Findings And Predictors. *Depression and Anxiety*, *31*, 964–971. doi:10.1002/da.22222

Muroff, J., Steketee, G., Himle, J., & Frost, R. (2010). Delivery of internet treatment for compulsive hoarding (D.I.T.C.H.). *Behaviour Research and Therapy*, *48*(1), 79–85. doi:10.1016/j.brat.2009.09.006

Muroff, J., Steketee, G., Rasmussen, J., Gibson, A., Bratiotis, C., & Sorrentino, C. (2009). Group Cognitive and Behavioural Treatment for Compulsive Hoarding: A Preliminary Trial. *Depression and Anxiety*, *26*(May), 634–640. doi:10.1002/da.20591

Muroff, J., Underwood, P., & Steketee, G. (2014). *Group Treatment for Hoarding Disorder: Therapist Guide (Treatments that work)*. New York: Oxford University Press.

Nedelisky, A., & Steele, M. (2009). Attachment to people and to objects in obsessive-compulsive disorder: an exploratory comparison of hoarders and non-hoarders. *Attachment & Human Development*, *11*(4), 365–83. doi:10.1080/14616730903016987

Nordsletten, A. E., Fernández de la Cruz, L., Billotti, D., & Mataix-Cols, D. (2013). Finders keepers: The features differentiating hoarding disorder from normative collecting. *Comprehensive Psychiatry*, *54*(3), 229–237. doi:10.1016/j.comppsych.2012.07.063

Nordsletten, A. E., Fernández de la Cruz, L., Pertusa, A., Reichenberg, A., Hatch, S. L., Mataix-Cols, D., … Mataix-Cols, D. (2013). The Structured Interview for Hoarding Disorder (SIHD): Development, usage and further validation. *Journal of Obsessive-Compulsive and Related Disorders*, *2*(3), 346–350. doi:10.1016/j.jocrd.2013.06.003

Nordsletten, A. E., Reichenberg, A., Hatch, S. L., Fernández De La Cruz, L., Pertusa, A., Hotopf, M., & Mataix-Cols, D. (2013). Epidemiology of hoarding disorder. *British Journal of Psychiatry*, *203*, 445–452. doi:10.1192/bjp.bp.113.130195

Onwumere, J., Bebbington, P., & Kuipers, E. (2011). Family interventions in early psychosis: specificity and effectiveness. *Epidemiology and Psychiatric Sciences*, *20*(02), 113–119. doi:10.1017/S2045796011000187

Orford, J., Templeton, L., Velleman, R., & Copello, A. (2005). Family members of relatives with alcohol, drug and gambling problems: a set of standardized questionnaires for assessing stress, coping and strain. *Addiction (Abingdon, England)*, *100*(11), 1611–24. doi:10.1111/j.1360-0443.2005.01178.x

Pekareva-Kochergina, A., & Frost, R. O. (2009). The Effects of a Biblio-Based Self-Help Program for Compulsive Hoarding. In *Paper presented at the Annual Meeting of the Association of Behavioral and Cognitive Therapies.* New York.

Pertusa, A., Frost, R. O., Fullana, M. a., Samuels, J., Steketee, G., Tolin, D., … Mataix-Cols, D. (2010). Refining the diagnostic boundaries of compulsive hoarding: A critical review. *Clinical Psychology Review*, *30*, 371–386. doi:10.1016/j.cpr.2010.01.007

Pertusa, A., Frost, R. O., & Mataix-Cols, D. (2010). When hoarding is a symptom of OCD: a case series and implications for DSM-V. *Behaviour Research and Therapy*, *48*(10), 1012–20. doi:10.1016/j.brat.2010.07.003

Pertusa, A., Fullana, M. A., Singh, S., Alonso, P., Menchón, J. M., & Mataix-Cols, D. (2008). Compulsive hoarding: OCD symptom, distinct clinical syndrome, or both? *The American Journal of Psychiatry*, *165*(10), 1289–98. doi:10.1176/appi.ajp.2008.07111730

Pharoah, F., Mari, J., Rathbone, J., & Wong, W. (2010). Family intervention for schizophrenia. *The Cochrane Database of Systematic Reviews*, (12), CD000088. doi:10.1002/14651858.CD000088.pub2

Pollock, L., Kellett, S. & Totterdell, P. (2014). An intensive time-series evaluation of the effectivenes of congitive behaviour therapy for hoarding disorder: A 2-year prospective study. *Psychotherapy Research 24(4), 485-495.* doi 10.1080/10503307.2013.843802

Prien, R. F., & Levine, J. (1984). Research and methodological issues for evaluating the therapeutic effectiveness of antidepressant drugs. *Psychopharmacology Bulletin*, *20*(2), 250–7. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/6374721

Reeves, B. C., Deeks, J. J., Higgins, J. P. T., & Wells, G. A. (2008). Chapter 13 : Including non-randomized studies. In J. P. T. Higgins & S. Green (Eds.), *Cochrane Handbook for Systematic Reviews of Interventions* (5.01 ed., pp. 1–34).

Rufer, M., Fricke, S., Moritz, S., Kloss, M., & Hand, I. (2006). Symptom dimensions in obsessive-compulsive disorder: prediction of cognitive-behavior therapy outcome. *Acta Psychiatrica Scandinavica*, *113*(5), 440–6. doi:10.1111/j.1600-0447.2005.00682.x

Sampson, J. M. (2013). The Lived Experience of Family Members of Persons Who Compulsively Hoard: A Qualitative Study. *Journal of Marital and Family Therapy*, *39*(3), 388–402. doi:10.1111/j.1752-0606.2012.00315.x

Sampson, J. M., Yeats, J. R., & Harris, S. M. (2012). An Evaluation of an Ambiguous Loss Based Psychoeducational Support Group for Family Members of Persons Who Hoard: A Pilot Study. *Contemporary Family Therapy*, *34*(4), 566–581. doi:10.1007/s10591-012-9214-6

Samuels, J. F., Bienvenu, O. J., Grados, M. A., Cullen, B., Riddle, M. A., Liang, K.-Y., … Nestadt, G. (2008). Prevalence and correlates of hoarding behavior in a community-based sample. *Behaviour Research and Therapy*, *46*(7), 836–44. doi:10.1016/j.brat.2008.04.004

Samuels, J. F., Bienvenu, O. J., Pinto, A., Fyer, A. J., McCracken, J. T., Rauch, S. L., … Nestadt, G. (2007). Hoarding in obsessive-compulsive disorder: results from the

OCD Collaborative Genetics Study. *Behaviour Research and Therapy*, *45*(4), 673–86. doi:10.1016/j.brat.2006.05.008

Saxena, S., Ayers, C. R., Dozier, M. E., & Maidment, K. M. (2015). The UCLA Hoarding Severity Scale: Development and validation. *Journal of Affective Disorders*, *175*, 488–93. doi:10.1016/j.jad.2015.01.030

Saxena, S., Ayers, C. R., Maidment, K. M., Vapnik, T., Wetherell, J. L., & Bystritsky, A. (2011). Quality of life and functional impairment in compulsive hoarding. *Journal of Psychiatric Research*, *45*(4), 475–80. doi:10.1016/j.jpsychires.2010.08.007

Saxena, S., Brody, A. L., Maidment, K. M., & Baxter, L. R. (2007). Paroxetine treatment of compulsive hoarding. *Journal of Psychiatric Research*, *41*(6), 481–487. doi:10.1016/j.jpsychires.2006.05.001

Saxena, S., & Maidment, K. M. (2004). Treatment of compulsive hoarding. *Journal of Clinical Psychology*, *60*(11), 1143–1154. doi:10.1002/jclp.20079

Saxena, S., Maidment, K. M., Vapnik, T., Golden, G., Rishwain, T., Rosen, R. M., & Bystritsky, A. (2002). Obsessive-Compulsive Hoarding: Symptom Severity and Response to Multimodal Treatment [CME]. *The Journal of Clinical Psychiatry*, *63*(1), 21–27. Retrieved from http://www-psychiatrist-com.libproxy.kcl.ac.uk/JCP/article/Pages/2002/v63n01/v63n0105.aspx

Saxena, S., & Sumner, J. (2014). Venlafaxine extended-release treatment of hoarding disorder. *International Clinical Psychopharmacology*, *29*, 266–273. doi:10.1097/YIC.0000000000000036

Schmalisch, C. S., Bratiotis, C., & Muroff, J. (2010). Processes in Group Cognitive and Behavioral Treatment for Hoarding. *Cognitive and Behavioral Practice*, *17*(4), 414–425. doi:10.1016/j.cbpra.2009.08.005

Schulz, K. F., Chalmers, I., Hayes, R. J., & Altman, D. G. (1995). Empirical evidence of bias. Dimensions of methodological quality associated with estimates of treatment effects in controlled trials. *JAMA : The Journal of the American Medical Association*, *273*, 408–412. doi:10.1097/00043764-199603000-00007

Scientific Brain Training PRO.. Retrieved from http://www.scientificbraintrainingpro.com/

Seedat, S., & Stein, D. J. (2002). Hoarding in obsessive-compulsive disorder and related disorders: a preliminary report of 15 cases. *Psychiatry and Clinical Neurosciences*, *56*(1), 17–23. doi:10.1046/j.1440-1819.2002.00926.x

Steer, R. A., & Beck, A. T. (1997). Beck Anxiety Inventory. In C. P. Zalaquett & R. J. Wood (Eds.), *Evaluating stress: A book of resources.* (pp. 23–40). Lanham, MD, US: Scarecrow Education.

Steketee, G., & Frost, R. (2003). Compulsive hoarding: Current status of the research. *Clinical Psychology Review*, *23*(7), 905–927. doi:10.1016/j.cpr.2003.08.002

Steketee, G., & Frost, R. O. (2007). *Compulsive Hoarding and Acquiring: Therapist Guide (Treatents That Work)*. New York: Oxford University Press.

Steketee, G., & Frost, R. O. (2013). *Treatment for Hoarding Disorder: Therapist Guide* (2nd ed.). New York: Oxford University Press.

Steketee, G., Frost, R. O., & Kyrios, M. (2003). Cognitive Aspects of Compulsive Hoarding. *Cognitive Therapy and Research*, *27*(4), 463–479. doi:10.1023/A:1025428631552

Steketee, G., Frost, R. O., Tolin, D. F., Rasmussen, J., & Brown, T. A. (2010). Waitlist-controlled trial of cognitive behavior therapy for hoarding disorder. *Depression and Anxiety*, *27*(5), 476–484. Retrieved from http://www.pubmedcentral.nih.gov/articlerender.fcgi?artid=2922687&tool=pmcentrez&rendertype=abstract

Steketee, G., Frost, R. O., Wincze, J., Greene, K. A. I., & Douglass, H. (2000). Group and Individual Treatment of Compulsive Hoarding: A Pilot Study. *Behavioural and Cognitive Psychotherapy*, *28*(1), 259–268.

Steketee, G., & Van Noppen, B. (2003). Family approaches to treatment for obsessive compulsive disorder. *Revista Brasileira de Psiquiatria*, *25*(1), 43–50. doi:10.1590/S1516-44462003000100009

Sterne, J. A. C., Gavaghan, D., & Egger, M. (2000). Publication and related bias in meta-analysis: Power of statistical tests and prevalence in the literature. *Journal of Clinical Epidemiology*, *53*(11), 1119–1129. doi:10.1016/S0895-4356(00)00242-0

Tarrier, N., & Wykes, T. (2004). Is there evidence that cognitive behaviour therapy is an effective treatment for schizophrenia? A cautious or cautionary tale? *Behaviour Research and Therapy*, *42*(12), 1377–1401. Retrieved from http://www.sciencedirect.com/science/article/pii/S0005796704001767

The Cochrane Collaboration. (2011a). *Cochrane Handbook for Systematic Reviews of Interventions*. (J. P. T. Higgins & S. Green, Eds.) (Version 5.). Retrieved from www.cochrane-handbook.org.

The Schizophrenia Commission. (2012). *The Abandoned Illness: A report by the Schizophrenia Commission*. Retrieved from http://www.schizophreniacommission.org.uk/the-report/

Timpano, K. R., Broman-Fulks, J. J., Glaesmer, H., Exner, C., Rief, W., Olatunji, B. O., … Schmidt, N. B. (2013). A taxometric exploration of the latent structure of hoarding. *Psychological Assessment*, *25*(1), 194–203. doi:10.1037/a0029966

Timpano, K. R., Çek, D., Fu, Z.-F., Tang, T., Wang, J.-P., & Chasson, G. S. (2015). A consideration of hoarding disorder symptoms in China. *Comprehensive Psychiatry*, *57*, 36–45. doi:10.1016/j.comppsych.2014.11.006

Timpano, K. R., Exner, C., Glaesmer, H., Reif, W., Keshavia, A., Brahler, E., & Wilhelm, S. (2011). The Epidemiology of the Proposed DSM-5 Hoarding Disorder: Exploration of the Acquisition Specifier, Associated Features, and Distress. *The Journal of Clinical Psychiatry*, *72*(6), 780–786. Retrieved from http://cat.inist.fr/?aModele=afficheN&cpsidt=24310573

Timpano, K. R., Keough, M. E., Traeger, L., & Schmidt, N. B. (2011). General life stress and hoarding: Examining the role of emotional tolerance. *International Journal of Cognitive Therapy*, *4*(3), 263–279. doi:10.1521/ijct.2011.4.3.263

Tolin, D. F. (2011). Challenges and advances in treating hoarding. *Journal of Clinical Psychology*, *67*(5), 451–5. doi:10.1002/jclp.20796

Tolin, D. F., Fitch, K. E., Frost, R. O., & Steketee, G. (2010). Family Informants' Perceptions of Insight in Compulsive Hoarding. *Cognitive Therapy and Research*, *34*(1), 69–81. doi:10.1007/s10608-008-9217-7

Tolin, D. F., Frost, R. O., & Steketee, G. (2007a). An Open Trial of Cognitive-Behavioural Therapy for Compulsive Hoarding. *Behaviour Research and Therapy*, *45*(7), 1461–1470.

Tolin, D. F., Frost, R. O., & Steketee, G. (2007b). *Buried in Treasures: Help for Compulsive Acquiring, Saving, and Hoarding*. New York: Oxford University Press.

Tolin, D. F., Frost, R. O., & Steketee, G. (2010). A brief interview for assessing compulsive hoarding: the Hoarding Rating Scale-Interview. *Psychiatry Research*, *178*(1), 147–52. doi:10.1016/j.psychres.2009.05.001

Tolin, D. F., Frost, R. O., Steketee, G., & Fitch, K. E. (2008). Family burden of compulsive hoarding: results of an internet survey. *Behaviour Research and Therapy*, *46*(3), 334–44. doi:10.1016/j.brat.2007.12.008

Tolin, D. F., Frost, R. O., Steketee, G., Gray, K. D., & Fitch, K. E. (2008). The economic and social burden of compulsive hoarding. *Psychiatry Research*, *160*(2), 200–11. doi:10.1016/j.psychres.2007.08.008

Tolin, D. F., Frost, R. O., Steketee, G., & Muroff, J. (2015). Cognitive Behavioral Therapy for Hoarding Disorder: a Meta-Analysis. *Depression and Anxiety*, *32*, 158–166. doi:10.1002/da.22327

Tolin, D. F., Meunier, S. a., Frost, R. O., & Steketee, G. (2010). Course of compulsive hoarding and its relationship to life events. *Depression and Anxiety*, *27*(9), 829–838. doi:10.1002/da.20684

Tolin, D. F., Stevens, M. C., Nave, A., Villavicencio, A. L., & Morrison, S. (2012). Neural mechanisms of cognitive behavioral therapy response in Hoarding Disorder : A pilot study. *Journal of Obsessive-Compulsive and Related Disorders*, *1*(3), 180–188. doi:10.1016/j.jocrd.2012.04.001

Tolin, D. F., Stevens, M. C., Villavicencio, A. L., Norberg, M. M., Calhoun, V. D., Frost, R. O., … Pearlson, G. D. (2012). Neural mechanisms of decision making in hoarding disorder. *Archives of General Psychiatry*, *69*(8), 832–41. doi:10.1001/archgenpsychiatry.2011.1980

Tolin, D. F., & Villavicencio, A. (2011). Inattention, but not OCD, predicts the core features of hoarding disorder. *Behaviour Research and Therapy*, *49*(2), 120–5. doi:10.1016/j.brat.2010.12.002

Tompkins, M. A. (2011). Working with families of people who hoard: a harm reduction approach. *Journal of Clinical Psychology*, *67*(5), 497–506. doi:10.1002/jclp.20797

Tompkins, M. A. (2015). *Clinician's Guide to Severe Hoarding*. New York, NY: Springer New York. doi:10.1007/978-1-4939-1432-6

Turner, K., Steketee, G., & Nauth, L. (2010). Treating Elders With Compulsive Hoarding: A Pilot Program. *Cognitive and Behavioral Practice*, *17*(4), 449–457. doi:10.1016/j.cbpra.2010.04.001

Twamley, E. W., Vella, L., Burton, C. Z., Heaton, R. K., & Jeste, D. V. (2012). Compensatory Cognitive Training for Psychosis. *The Journal of Clinical Psychiatry*, *73*(09), 1212–1219. doi:10.4088/JCP.12m07686

Van der Gaag, M., Smit, F., Bechdolf, A., French, P., Linszen, D. H., Yung, A. R., … Cuijpers, P. (2013). Preventing a first episode of psychosis: meta-analysis of randomized controlled prevention trials of 12 month and longer-term follow-ups. *Schizophrenia Research*, *149*(1-3), 56–62. doi:10.1016/j.schres.2013.07.004

Whitfield, G. (2003). The evidence base for cognitive-behavioural therapy in depression: delivery in busy clinical settings. *Advances in Psychiatric Treatment*, *9*(1), 21–30. doi:10.1192/apt.9.1.21

Wilbram, M., Kellett, S., & Beail, N. (2008). Compulsive hoarding: a qualitative investigation of partner and carer perspectives. *The British Journal of Clinical Psychology / the British Psychological Society*, *47*(Pt 1), 59–73. doi:10.1348/014466507X240740

Woody, S. R., Kellman-McFarlane, K., & Welsted, A. (2014). Review of cognitive performance in hoarding disorder. *Clinical Psychology Review*, *34*(4), 324–336. doi:10.1016/j.cpr.2014.04.002

Wykes, T., Huddy, V., Cellard, C., McGurk, S. R., & Czobor, P. (2011). A meta-analysis of cognitive remediation for schizophrenia: methodology and effect sizes. *The American Journal of Psychiatry*, *168*(5), 472–85. doi:10.1176/appi.ajp.2010.10060855

Wykes, T., Steel, C., Everitt, B., & Tarrier, N. (2008). Cognitive behavior therapy for schizophrenia: effect sizes, clinical models, and methodological rigor. *Schizophrenia Bulletin*, *34*(3), 523–37. doi:10.1093/schbul/sbm114

Yalom, I. D., & Leszcz, M. (2005). *Theory and Practice of Group Psychotherapy* (Fifth.). Basic Books.

Yesufu-Udechuku, a., Harrison, B., Mayo-Wilson, E., Young, N., Woodhams, P., Shiers, D., … Kendall, T. (2015). Interventions to improve the experience of caring for people with severe mental illness: systematic review and meta-analysis. *The British Journal of Psychiatry*, *206*(4), 268–274. doi:10.1192/bjp.bp.114.147561

Zigmond, A. S., & Snaith, R. P. (1983). The Hospital Anxiety and Depression Scale. *Acta Psychiatrica Scandinavica*, *67*(6), 361–70. doi:10.1111/j.1600-0447.1983.tb09716.x

Zohar, J., & Westenberg, H. G. (2000). Anxiety disorders: a review of tricyclic antidepressants and selective serotonin reuptake inhibitors. *Acta Psychiatrica Scandinavica. Supplementum*, *403*(6), 39–49. doi:10.1111/j.1600-0447.2000.tb10947.x

6. Appendices

Appendix 1: Diagnostic criteria for Hoarding Disorder (from DSM-5; APA, 2013)

A. Persistent difficulty discarding or parting with possessions, regardless of their actual value.

B. This difficulty is due to a perceived need to save the items and distress associated with discarding them

C. The symptoms result in the accumulation of possessions that congest and clutter active living areas and substantially compromise their intended use. If living areas are uncluttered, it is only because of the interventions of third parties (e.g. family members, cleaners, authorities)

D. The hoarding causes clinically significant distress or impairment in social, occupational or other important areas of functioning (including maintaining a safe environment for self and others).

E. The hoarding is not attributable to another medical condition (e.g. brain injury, cerebrovascular disease, Prader–Willi syndrome).

F. The hoarding is not better accounted for by the symptoms of another DSM-5 disorder (e.g. hoarding due to obsessions in obsessive–compulsive disorder, decreased energy in major depressive disorder, delusions in schizophrenia or another psychotic disorder, cognitive deficits in dementia, restricted interests in autism spectrum disorder).

Appendix 2: Data extraction form

Author	
Year	
Location	
Population (HD or relatives)	
Purpose of study	
N recruited	
Dropout rate N	
Age M (SD)	
Gender	
Ethnicity	
Study Design	
Recruitment method	
Allocation/randomisation and method	
Control group	
Type of treatment	
Number of treatment sessions	
Treatment adequately described	
Who delivered treatment	
Was a protocol used	
Was adherence to treatment assessed	
Definition of HD for inclusion	
HD measures used	
Outcome on HD symptoms	
Effect size (if reported)	
Clinically significant change and how defined?	
Secondary outcome measures used (of impact on life/distress, reductions of co morbid psychiatric symptoms (mood,	

anxiety)	
Secondary outcomes	
Assessment period	
Who conducted assessments?	
Blinding?	
Analysis including how handled missing data, Power calculation	
Summary/Study conclusions	

Appendix 3: The Clinical Trials Assessment Measure (Tarrier & Wykes, 2004)

Sample—two questions: maximum score = 10

Q1: is the sample a convenience sample (score 2) or a geographic cohort (score 5), highly selective sample, e.g., volunteers (score 0)

Convenience sample—e.g., clinic attenders, referred patients or Geographic cohort—all patients eligible in a particular area

Q2: is the sample size greater than 27 participants in each treatment group (score 5) or based on described and adequate power calculations (score 5)

Allocation—three questions: maximum score = 16

Q3: is there true random allocation or minimisation allocation to treatment groups (if yes score 10)

Q4: is the process of randomisation described (score 3)

Q5: is the process of randomisation carried out independently from the trial research team (score 3)

Assessment (for the main outcome)—five questions: maximum score = 32

Q6: are the assessments carried out by independent assessors and not therapists (score 10)

Q7: are standardised assessments used to measure symptoms in a standard way (score 6), idiosyncratic assessments of symptoms (score 3)

Q8: are assessments carried out blind (masked) to treatment group allocation (score 10)

Q9: are the methods of rater blinding adequately described (score 3)

Q10: is rater blinding verified (score 3)

Control groups—one question: maximum score = 16

Q11: TAU is a control group (score 6) and/or a control group that controls for non-specific effects or other established or credible treatment (score 10)

Analysis—two questions: maximum score = 15

Q12: the analysis is appropriate to the design and the type of outcome measure (score 5)

Q13: the analysis includes all those participants as randomised (sometimes referred to as an intention to treat analysis) (score 6) and an adequate investigation and handling of drop outs from assessment if the attrition rate exceeds 15% (score 4)

Active treatment—three questions: maximum score = 11

Q14: was the treatment adequately described (score 3) and was a treatment protocol or manual used (score 3)

Q15: was adherence to the treatment protocol or treatment quality assessed (score 5) where the criterion is not reached for any question score = 0

Total score: maximum score = 100

Appendix 4: *Table 3 – Mean scores in each domain of CTAM*

Domain of CTAM	Question	Maximum possible score	Average score (SD)
1. Sampling	*Recruitment method*	5	0.71 (0.99)
	Sample size	5	1.18 (2.19)
2. Allocation	*Randomisation*	10	2.35 (4.37)
	Randomisation described	3	0.18 (0.73)
	Independent randomisation	3	0 (0)
3. Assessment	*Independent assessments*	10	2.35 (4.37)
	Standardised measures	6	6 (0)
	Blinding	10	1.76 (3.93)
	Blinding described	3	0 (0)
	Blinding verified	3	0 (0)
4. Control group	*TAU or controls for non-specific effects*	16	1.18 (3.32)
5. Analysis	*Analysis appropriate to design*	5	5 (0)
	Analysis includes all participants as randomised and appropriate handling of dropouts	10	3.53 (3.04)
6. Active treatment	*Treatment described and manual/protocol used*	6	4.76 (1.52)
	Adherence to protocol assessed	5	0.59 (1.17)

Appendix 5: *Table 4 – Summary table of significant effects of intervention studies*

Study (First Author, year)	Intervention type	Significant effect on HD symptoms (either SI-R/HRS/UHSS)	Significant impact on functional impairment	Significant impact on anxiety or depression
Ayers et al. (2011)	Individual CBT	+	O	+
Steketee et al.(2010)	Individual CBT	+	n/a	O
Tolin et al. (2007)	Individual CBT	+	n/a	n/a
Turner et al. (2010)	Individual CBT	O (improvement on CIR only)	+	n/a
Gilliam et al. (2011)	Group CBT	+	+	+
Tolin et al. (2012)	Group CBT	O	n/a	n/a
Muroff et al. (2012)	Group CBT	+	+	+
Muroff et al. (2009)	Group CBT	+	n/a	+
Frost et al. (2011) Study 1	CBT bibliotherapy based support group	+	+	O
Frost et al. (2011) Study 2	CBT bibliotherapy based support group	+	+	O
Frost et al. (2012)	CBT bibliotherapy based support group	+	+	n/a
Saxena et al. (2007)	Pharmacotherapy	+	n/a	+

Saxena & Sumner (2014)	Pharmacotherapy	+	n/a	+
Ayers et al. (2014)	Cognitive remediation + behavioural therapy	+	n/a	n/a
Dimauro et al. (2014)	Cognitive remediation	O	n/a	n/a
Chasson et al. (2014)	Relatives only	O	n/a	n/a
Muroff et al. (2010)	Online CBT/Self-help	+	n/a	n/a

CIR=Clutter Image Rating scale, HRS=Hoarding Rating Scale, SI-R=Saving Inventory Revised, UHSS=UCLA Hoarding Severity Scale

n/a=not measured in the study, O=no significant change, += positive effect

Institute of Psychiatry, Psychology and Neuroscience

King's College London

May 2015

Main Research Project

The impact of caregiving in Hoarding Disorder: piloting a brief psychoeducational group for relatives of hoarders.

Supervised by:

Dr Juliana Onwumere

Dr Lorena Fernández de la Cruz

Professor David Mataix-Cols

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Abstract:
Hoarding Disorder (HD) is newly recognised mental health disorder, commonly associated with substantial functional disability, which represents a major public health burden. It is becoming increasingly clear that hoarding problems can also have a major impact on the relatives (carers) of people who hoard. This study aimed to: (1) identify the factors associated with distress and burden in carers of people with HD and (2) evaluate the impact of a brief cognitive-behaviour therapy (CBT) based intervention for this group focusing on understanding HD, wellbeing, burden, and coping efforts. Fifty-three relatives took part in the first phase of the study, and twelve carers completed the CBT-based intervention. Results indicated that carer burden and distress were associated with negative appraisals of HD and expressed emotion. The group intervention impacted positively on carer wellbeing, understanding of HD and positive experiences of caregiving. These gains were maintained after one month. HD can have a substantial negative effect on families and carer-based interventions show promise in improving carer experience and are acceptable to carers. Future studies incorporating a randomised controlled design are therefore indicated.

1. Introduction

1.1 Hoarding Disorder

Hoarding disorder (HD) is a severe and disabling mental health condition, which can have a far-reaching impact on the sufferers, their families and wider society (Frost, Steketee, & Williams, 2000; Saxena et al., 2011; Tolin, Frost, Steketee, & Fitch, 2008; Tolin, Frost, Steketee, Gray, & Fitch, 2008). HD has been included as a condition in its own right in the recently updated edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5, American Psychiatric Association, 2013), whereas previously hoarding behaviour was thought to be a symptom of other disorders (obsessive compulsive disorder (OCD) or obsessive compulsive personality disorder (OCPD)). The primary symptom of HD is the persistent difficulty parting with possessions, regardless of their value. People with HD report the difficulty discarding is due to a 'need to save' possessions and experiencing distress when attempting to discard possessions. The avoidance of discarding leads to an excessive build-up of clutter within the home which, at clinical levels, causes problems using rooms for their intended purpose (e.g. difficulty using kitchen to prepare food) (full DSM-5 criteria are detailed in Appendix 1). HD was once thought to be a rare condition, but epidemiological research suggests prevalence rates to fall between 1.5% and 5% of the adult population (Iervolino et al., 2009; Mueller, Mitchell, Crosby, Glaesmer, & de Zwaan, 2009; Nordsletten et al., 2013; Samuels et al., 2008; Timpano et al., 2011).

HD is associated with substantial disability and impairment in various areas of functioning. Hoarding patients show higher levels of social and family dysfunction than controls or OCD patients (Frost, Steketee, Williams, & Warren, 2000) and reduced quality of life compared to healthy controls (Saxena et al., 2011). People with HD generally have worse than average physical health status and are more likely to be unemployed, single, separated or divorced than comparison groups (Mataix-Cols, Billotti, Fernández de la Cruz, & Nordsletten, 2013; Nordsletten et al., 2013; Tolin, Frost, Steketee, & Fitch, 2008; Tolin, Frost, Steketee, Gray, & Fitch, 2008). Up to 50% of people with HD also have at least one comorbid psychiatric condition such as depression and anxiety (Frost, Steketee, & Tolin, 2011). Hoarding can pose a more direct threat to physical health and safety. The risk of fire or falls due to clutter for the person with HD, their cohabitants and neighbours are high,

particularly for the elderly (Kim, Steketee, & Frost, 2001). In one U.S. study (Frost, Steketee, & Williams, 2000), 64% of health officials surveyed had received complaints about hoarding, usually from neighbours, fire services and police departments. Social service involvement is also not uncommon, with as many as one in 25 people with HD having had a dependent child, an elderly relative or pet removed from the home due to poor living conditions (Tolin, Frost, Steketee, Gray, et al., 2008). The costs to society are high in terms of health and social service provision, including the direct costs of house clear-outs. For example, recent figures from the USA suggest that the average amount spent on clutter removal is over £2,000 per case (McGuire, Kaercher, Park, & Storch, 2013).

1.2 The impact of Hoarding Disorder on the family

It is increasingly recognised that, as well as the adverse impact on the individual and society, HD can have severe implications for family members and relationships. Growing up with a parent or other family member with HD can be associated with a range of negative effects, including reports of embarrassment, feeling unable to have friends and other family around to the home, and arguments regarding the clutter and hoarding. Büscher, Dyson, and Cowdell (2014) reviewed the main studies looking at the impact of HD on families using thematic analysis (Braun & Clarke, 2008) and identified 3 overarching themes: “quality of life”, “shattered families”, and “rallying around”. The deleterious effect of HD on the wellbeing of family members has been established in several studies. The results of a large scale internet survey suggested that growing up in a hoarded home was associated with greater childhood distress and family strain (Tolin, Frost, Steketee, & Fitch, 2008). Drury and colleagues studied the wellbeing of people with HD and their relatives, compared to non-clinical collectors, and found that the level of functional impairment experienced by relatives of people with HD was equivalent to that of the person with HD themselves (Drury, Ajmi, Fernández de la Cruz, Nordsletten, & Mataix-Cols, 2014). Furthermore, relatives reported poorer emotional wellbeing and greater role limitations due to emotional problems compared to relatives of collectors.

HD can have a major impact on the quality of relationships between family members. Almost two thirds of people with hoarding difficulties are able to identify

relationship problems with family members due to their hoarding (Frost & Gross, 1993) and Tolin et al. (2008) found high levels of rejection of the hoarding relative in family members. Poorer relationships have also been reported in qualitative studies, often attributed to the hoarding itself (Sampson, 2013; Wilbram, Kellett, & Beail, 2008). Participants describe anger and frustration toward the relative with HD, high levels of conflict and a sense of loss in relation to family life, loss of a 'normal' home and relationship with the person. In some cases relationships had even broken down completely due to the hoarding (Sampson, 2013; Wilbram et al., 2008). Park, Lewin, and Storch (2014) used path analysis to look at influences on the quality of parent-offspring relationships in HD. They found that as hoarding severity increased, family relationships became more dysfunctional and this in turn negatively impacted parent-offspring relationships.

The third theme identified by Büscher and colleagues (2014) was "rallying around", which included families supporting the individual with HD and 'family accommodation' to the hoarding behaviours. Families often play a major supporting role for people with HD who may be otherwise quite socially isolated (e.g. Kim et al., 2001; Pertusa et al., 2008). The concept of family accommodation has been much studied in OCD (e.g. Lebowitz, Panza, Su, & Bloch, 2012) and eating disorders (Sepulveda, Kyriacou, & Treasure, 2009; Treasure et al., 2008). Family accommodation refers to the process where families inadvertently become part of the dysfunctional behaviour which maintains the condition and contributes to patient and family distress (Amir, Freshman, & Foa, 2000; Ramos-Cerqueira, Torres, Torresan, Negreiros, & Vitorino, 2008; Sepulveda et al., 2009; Storch, Geffken, Merlo, Jacob, et al., 2007). Family accommodation can be targeted successfully in interventions (e.g. Goddard et al., 2011; Storch, Geffken, Merlo, Mann, et al., 2007). Accommodation has been found in HD families in various forms, such as not discarding acquired items in order to avoid arguments with the person, purchasing storage facilities, or even storing possessions at their own homes when space becomes limited in the hoarding relatives' home (Drury et al., 2014; Nordsletten et al., 2014; Park et al., 2014). Park et al. (2014) found accommodation to be significantly associated with functional impairment in children of people with HD.

As well as the hoarding behaviour and presence of clutter, the lack of available information about HD and misattribution of it as a problem of laziness or messiness can be a major source of frustration for family members (Marley, Thompson, & Onwumere, 2015, unpublished thesis). Similarly, misunderstanding within the general public can further add to the stigma and embarrassment reported by families (Wilbram et al., 2008). Often individuals with HD fail to recognise their symptoms as problematic, which can be a source of family conflict (Drury et al., 2014; Frost, Steketee, Youngren, & Mallya, 1999; Tolin, Fitch, Frost, & Steketee, 2010). Poor insight also contributes to low rates of treatment seeking in people with hoarding problems (Damecour & Charron, 1998), potentially adding to the distress of family members. In general, people with HD have been reported to have more interpersonal problems than controls, which could be as a result of the hoarding and/or certain personality characteristics often seen in people with HD (Frost et al., 2000; Grisham, Steketee, & Frost, 2008; Medard & Kellett, 2014).

1.3 Understanding the experience of caregivers

Our understanding of the impact of HD on family members can be informed by extensive and well-established literature on family experiences in other severe and enduring mental health conditions such as schizophrenia spectrum disorders (i.e., psychosis), which may show some parallels to HD in terms of the challenges faced by family members. Similarities can be drawn between HD and psychosis in terms of the course of the illnesses; both of which start at a relatively young age, are usually enduring, and are often characterised by low levels of insight, contributing to poorer engagement in treatment. However, there are also differences between the conditions which mean that the two disorders may affect families differently. One difference is the presence of fluctuating symptoms in psychosis; i.e. 'crises' are more common than in HD, where presentation is typically more chronic. Another difference is in the presentation of the individual; people with psychosis may behave in an unusual manner which is noticeable to others, whereas HD can be undetectable in a person's behaviour outside of the home, and remain a 'secret' from others.

The literature often refers to 'caregivers' or 'carers', meaning people who provide informal care or support to the person with a mental health problem, which can include family members, partners, and close friends. The critically important role played by informal carers in mental health conditions is increasingly recognised, including the financial savings for the economy. For instance, in England alone, informal carers are estimated to save £615 million a year on the costs associated with psychosis (Mangalore & Knapp, 2007). The negative consequences of caregiving are often referred to as 'caregiver burden' (Awad & Voruganti, 2008), although positive and rewarding experiences of caregiving such as satisfaction and increased understanding have also been recognised as important, and could potentially negate some of the negative effects of caregiving (Chen & Greenberg, 2004; Kulhara, 2012; Szmukler et al., 1996).

Negative aspects of caregiving found in mental health conditions are numerous and include feelings of loss, grief (Miller, Dworkin, Ward, & Barone, 1990; Patterson, Birchwood, & Cochrane, 2005), anger, guilt and shame (e.g. Jones, 2004), and stigma (e.g. Magaña, Ramírez García, Hernández, & Cortez, 2007; Mak & Cheung, 2012; Wahl & Harman, 1989). Caregiving, independent of the health condition, is linked to comorbid mental health conditions and poor psychological health, particularly in those providing more than 20 hours of care per week (Smith et al., 2014; Stansfeld et al., 2014). Approximately 40% of carers for individuals with psychosis report clinical levels of depression and anxiety (Jansen, Gleeson, & Cotton, 2015; Kuipers & Raune, 2000). Similar reports also been described in caregivers of people with OCD (Kalra, Kamath, Trivedi, & Janca, 2008; Magliano, Tosini, Guarneri, Marasco, & Catapano, 1996; Ramos-Cerqueira et al., 2008). Studies in psychosis have identified the true cost of caregiving in terms of financial (up to £32 million in loss of productivity; Mangalore & Knapp 2007) and functional impairment, and poorer mental and physical health in carers (e.g. Gutiérrez-Maldonado, Caqueo-Urízar, & Kavanagh, 2005; Perlick, Hohenstein, Clarkin, Kaczynski, & Rosenheck, 2005).

Negative appraisals of psychosis (e.g. regarding its severity, timeline and responsiveness to treatment) have been linked to increased levels of caregiver burden and distress (e.g. Barrowclough, Lobban, Hatton, & Quinn, 2001; Fortune,

Smith, & Garvey, 2005; Lobban, Barrowclough, & Jones, 2005; Onwumere et al., 2008). For example, in Fortune et al. (2005), carers of adults with psychosis who viewed their relative's illness as chronic, with severe consequences and who did not think treatment would be very effective, but did think that their relative could exert control over their illness, reported higher levels of distress. This finding may be relevant for HD where information on the condition and availability of effective treatments are limited.

Another predictor of caregiver burden in other disorders is expressed emotion (EE). EE has been described and defined in several ways, but in its simplest form reflects the reported attitude and behaviours expressed by a caregiver toward the person they care for and can be considered a proxy of the quality of a caregiving relationship. Ratings of EE can be classified as low or high; high EE are those scoring above threshold on levels of criticism/hostility and/or emotional over involvement. High EE is found more commonly in relationships where one person has mental health condition compared to healthy controls (e.g. Kyriacou, Treasure, & Schmidt, 2008). High EE carers report greater levels of emotional distress and burden than low EE carers (Barrowclough & Parle, 1997; Raune, Kuipers, & Bebbington, 2004; Scazufca & Kuipers, 1996). High EE can be predictive of poorer outcomes in psychosis, in terms of a greater number of relapses and readmissions (Bebbington & Kuipers, 1994; Butzlaff & Hooley, 1998; Cechnicki, Bielańska, Hanuszkiewicz, & Daren, 2013). In OCD, high EE also predicts poorer outcomes in therapy (Chambless & Steketee, 1999), a greater chance of relapse, and is associated with increased burden and lower quality of life in carers (Cherian, Pandian, Bada Math, Kandavel, & Janardhan Reddy, 2014). In psychosis, EE is also linked to less adaptive coping styles, such as avoidant coping (Raune et al., 2004), which are themselves related to increased care distress and burden (Budd, Oles, & Hughes, 1998; Fortune et al., 2005; Onwumere et al., 2011).

Kuipers, Onwumere and Bebbington (2010) proposed a cognitive model of caregiving responses in psychosis, which integrated the concepts of illness appraisals, caregiving experiences, quality of the relationship (EE), coping, and social support. The model proposed that the cognitive and emotional reactions of the carer

to the illness depend on how they appraise the illness and their loved one's behaviour. These appraisals are influenced by the nature of the relationship prior to onset of the illness. For example if the relationship was previously positive, then the episode of psychosis may be appraised as 'not their fault' and 'needing support', resulting in sadness but also helpful action to support the person. This relationship will be associated with low EE behaviours and seeking out appropriate social support. The model also provided guidance on types of interventions likely to be helpful for different types of reactions to caregiving. The model raises interesting questions when applied to HD, where the hoarding has potentially been present for the entire duration of the relationship between the person and their carer.

1.4 Carer interventions

To date, the main research efforts in the treatment of HD have focused almost exclusively on the person who hoards. However, improvement in HD symptoms has proved to be limited thus far. Data from a recent meta-analysis of cognitive-behavioural therapy for HD (Tolin, Frost, Steketee, & Muroff, 2015) showed that on average, only 35% of patients demonstrated clinically significant change, despite lengthy and intensive treatment. There was also little impact of the interventions on quality of life and functional impairment. We know that many people with HD do not seek treatment (Tolin, Frost, Steketee, Gray, et al., 2008), under-report their symptoms (Dimauro, Tolin, Frost, & Steketee, 2013) and often drop out of treatment (e.g. Gilliam et al., 2011; Tolin, Frost, & Steketee, 2007; Turner, Steketee, & Nauth, 2010). Families can be instrumental in encouraging relatives to seek help and family based interventions are a well-established therapeutic approach employed successfully with other severe mental health conditions (e.g., psychosis, bipolar disorder, OCD) (Miklowitz, 2006; Pharoah, Mari, Rathbone, & Wong, 2010; Renshaw, Steketee, & Chambless, 2005), including those at the very early stages of illness (Bird et al., 2010; Miklowitz et al., 2014). Family interventions are efficacious in reducing rates of patient relapse and improving their social functioning and treatment adherence (Bird et al., 2010; Cuijpers, 1999; Pharoah et al., 2010). They are cost effective and in psychosis can positively impact on carer burden and readiness to continue providing care (Berglund, Vahlne, & Edman, 2003; Christenson, Crane, Bell, Beer, & Hillin, 2014; Lobban et al., 2013; NICE, 2014).

Family interventions are indicated treatments in HD, where they could potentially augment outcomes for the hoarding relative, reach less motivated individuals and impact on carer only outcomes such as wellbeing and coping styles. Carer-only interventions also have the potential to impact on caregiving experiences and improve the relationship quality through targeting specific carer appraisals about the illness, caregiving experiences, and coping. Previous research has suggested that family members of people with HD could benefit from education and support interventions to facilitate their understanding of and adaptive coping with the condition, as well as being able to provide optimal support to their relative (Büscher et al., 2014; Chasson, Carpenter, Ewing, Gibby, & Lee, 2014; Sampson, Yeats, & Harris, 2012; Tompkins, 2011; Wilbram et al., 2008).

However, interventions involving carers of people with HD are novel and sparsely explored. Two case reports have described promising outcomes from interventions for HD involving family members. Tompkins (2011) described a 'harm-reduction' (Marlatt, 1998) approach for families of people with HD and low motivation to seek help. Their harm reduction protocol aims to improve problematic relationships in order to motivate the person with HD to accept help and emphasises reducing potential harm rather than decluttering per se. They reported two cases where there was positive improvement in clutter and the motivation of the person with HD to seek help post-intervention, although no outcome measures were reported. Ale, Arnold, Whiteside and Storch (2013) presented a case study of family based CBT for a child with hoarding behaviour, with reductions in scores on the Child Saving Inventory (a measure of hoarding symptoms, Storch et al., 2011) and OCD symptoms following treatment.

Two small-scale U.S. studies have evaluated carer-only interventions in HD. Sampson et al. (2012) piloted a six week psychoeducational-style support group for eight carers. The authors did not report any quantitative outcomes, but six participants completed qualitative interviews post intervention. The results highlighted subjective improvements to participants' knowledge and understanding of hoarding, levels of distress, and positive interactions with their relative (Sampson et al., 2012). Chasson et al. (2014) reported the results of a fourteen week 'family-as-motivators'

intervention package for nine carers. The package included modules on psychoeducation, harm reduction, motivational interviewing training, and family accommodation prevention. Results suggested improvements in the use of helpful coping strategies, hopefulness, use of motivational interviewing strategies, and understanding of hoarding. There were some notable improvements in the impact of HD on the family from pre-to-mid training, but no significant change on actual HD symptoms as reported by carers. However, four of the nine carers (44%) dropped out before the end of treatment, which left the end sample size of five carers and consequently limited the interpretation of the results. The authors suggested one reason for dropout may have been the long duration of the intervention. Aside from small sample sizes, another limitation of these previous studies is that participants were recruited through mental health service referrals, which is unlikely to be representative of many carers whose relatives are ambivalent about seeking help and may not be in contact with mental health services regarding their hoarding. In addition, problematic hoarding in the relative was assessed only via a brief screening measure (the Hoarding Rating Scale; Tolin, Frost, & Steketee, 2010), and thus it is unclear to what extent relatives met full HD criteria.

It seems clear that further testing of carer interventions in HD is warranted, which should take account of recommendations from previous studies (i.e. inclusion of psychoeducational material, small group setting; Sampson et al., 2012). Given that HD is now recognised as a mental health condition, and the evidence base reviewed here regarding the needs of carers, including the need for information (Sampson, 2013; Wilbram et al., 2008), interventions should be designed to support and inform carers about HD, facilitate the uptake and implementation of adaptive coping strategies and address common issues faced by carers of people who hoard. Brief carer-based psycho-education groups have been increasingly studied in psychosis (Chien & Norman, 2009; Gutiérrez-Maldonado, Caqueo-Urízar, & Ferrer-García, 2009; Sin & Norman, 2013) and linked to positive outcomes such as increased knowledge about the condition, improved coping and quality of life and reductions in distress (Lowenstein, Butler, & Ashcroft, 2010; Petrakis, Oxley, & Bloom, 2012; Sin & Norman, 2013; Yesufu-Udechuku et al., 2015). The emerging evidence on carer

interventions in HD also indicates the importance of psychoeducational elements, along with a supportive group environment and a relatively brief length of intervention (Chasson et al., 2014; Sampson et al., 2012).

1.5 Rationale for current study

The aims of the present study were twofold. The experiences of relatives ('carers') of people with HD are not yet fully understood in terms of caregiver burden, relationship with the person who hoards, appraisals of the illness, ways of coping, wellbeing, and emotional functioning. Thus, Phase 1 of this study aims to investigate the experiences of carers using validated measures from the caregiving literature. It will compare outcomes in terms of distress and caregiver burden with existing data on carers of people with psychosis, a better studied group of carers, and with whom similarities between conditions have been drawn. The second part of the study (Phase 2), comprises a proof of concept study of a brief CBT psychoeducational group intervention for carers of people with HD, employing quantitative measures to determine the effects of the group on carer understanding of HD and levels of wellbeing, burden, and coping efforts.

1.6 Hypotheses

Phase 1:

1. Carer's negative appraisals regarding the relative's HD will be associated with higher levels of carer burden and psychological distress. Negative appraisals of the relative's illness and behaviour will be associated with higher levels of carer burden and distress.
2. There will be a positive association between EE levels, distress, and poorer coping in carers of people with HD.
3. Carers of people with HD will show equivalent or higher levels of caregiving burden and distress than carers of people with psychosis.

Phase 2:

1. Carers' understanding of hoarding, including causality and symptomatology, will improve post intervention.
2. Carer reports of wellbeing, distress, and positive aspects of caregiving will improve post intervention.

3. The intervention will improve adaptive coping.
4. The intervention will be deemed acceptable by participants.

2. Methods

The study was approved by King's College London's Research Ethics Committee (reference PNM/13/14-28, see appendix 2).

2.1 Sample

The sample consisted of carers of people who met DSM-5 criteria for HD. A 'carer' was defined as: a parent, relative, spouse, or partner of an individual with likely HD (as according to the Structured Interview for Hoarding Disorder (SIHD; Nordsletten et al., 2013, administered with the carer). The inclusion criteria were:

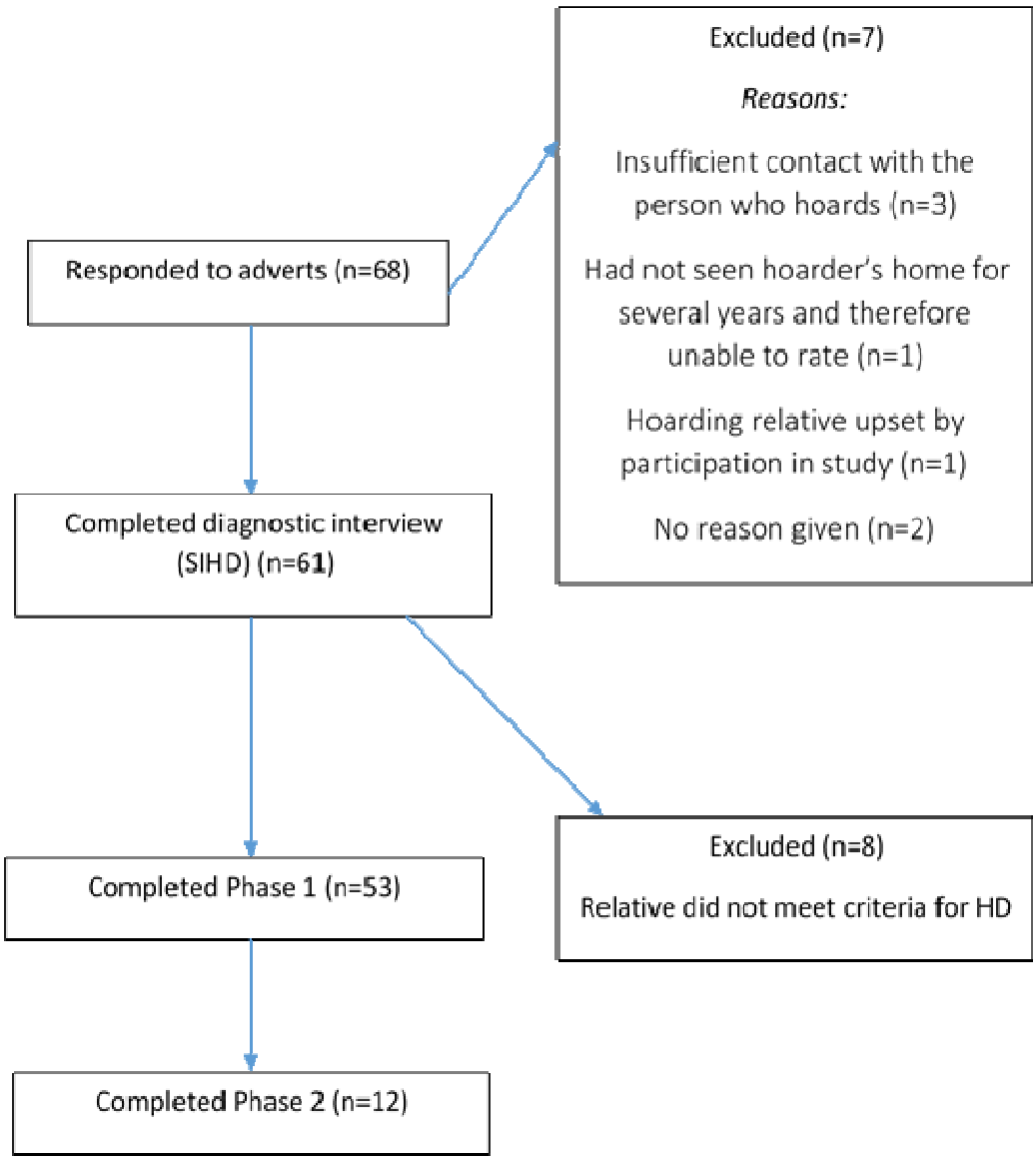
a) age 18 or over;
b) being a carer of a person with likely HD
c) regular weekly contact with the individual, including telephone contact. Similar definitions of carers have been employed in previous research with carers from other mental health conditions (e.g. Kuipers et al., 2006).
d) (Phase 2 only) Able to travel to London for group sessions.

Exclusion criteria for the study were:

a) insufficient command of English to complete measures and engage in group intervention;
b) meeting criteria for HD themselves (screened with Hoarding Rating Scale; Tolin, Frost, et al., 2010).

The participant flow chart is detailed in Figure 1 and recruitment procedure detailed in the section below. Fifty-three carers participated in Phase 1 and twelve in Phase 2.

Figure 1: Recruitment flow chart



2.2 Recruitment procedure

Participants were recruited through advertisements placed on self-help forums for hoarding (e.g. helpforhoarders.co.uk; childrenofhoarders.com) and the research team's website (hoardingstudy.com). Additionally, carers of people meeting criteria for HD from an existing database who had participated in previous studies or had contacted the Hoarding research team at KCL were invited to take part. All carers who contacted the team were sent an information sheet (see Appendix 3) and were screened using the Hoarding Rating Scale Interview (HRS-I; Tolin, Frost, et al., 2010).

Following this they signed a consent form and completed the interview assessments over the phone and self-report measures via online or paper questionnaires.

2.3 Design

2.3.1. Phase 1

Phase 1 was a baseline, cross-sectional study. In order to contextualise outcomes, distress and burden levels were compared to a group of carers of people with psychosis (n=86), data for whom were collected as part of a trial for individual and family therapies in psychosis (Garety et al., 2008). Relevant data (psychological distress and experiences of caregiving) were extracted from the original study database. Distress data was available from 69 psychosis carers, and experiences of caregiving data from 63.

2.3.2. Phase 2

Phase 2 was a pilot investigation of a brief (6 weekly sessions + 1 month follow-up session), standardized psychoeducational group intervention for carers of people with HD. The groups were held in a university building for two hours per session on a weekday evening. Group facilitators were a senior clinical psychologist with specialism in carer interventions (JO), a research clinical psychologist with specialism in HD (LFC) and the first author, a trainee clinical psychologist. Two groups were conducted with six participants in each group. Participants completed measures at baseline, post-intervention and 1 month follow-up. As part of a separate study a subset of participants completed interviews regarding their experience of the group (Khan, Thompson and Onwumere, 2015, unpublished thesis).

Briefly, the intervention protocol existed of:

Session 1: Emotional processing and opportunity to 'tell their story'

Sessions 2-3: Psychoeducation about HD including causes, symptoms and treatments.

Sessions 4-5: Problem solving common difficulties faced by carers of people with HD.

Session 6: Promoting carer wellbeing.

A more detailed description of the intervention protocol is included in Appendix 4.

2.4 Assessment measures

Assessment measures are included in Appendix 5.

Social demographic information (Appendix 5.1) was collected from all participants including gender, age, ethnicity, amount of contact with the person who hoards, and their relationship to the person. Carers also provided details on the age and gender of their relative and estimate of illness length. To determine appraisals of current social support a single question was asked "In general, do you feel that you have someone that you can confide in?".

2.4.1. Hoarding measures

All measures were self-report, other than the diagnostic interview (SIHD) and the Five Minute Speech Sample, which were conducted over the phone.

The *SIHD* (Nordsletten et al., 2013, Appendix 5.2), a semi-structured clinical interview for HD, was used with the carer to establish whether the hoarding relative would meet criteria for HD. Relative ratings of HD have been shown to correlate highly with self-report from hoarders (Drury et al., 2014). The SIHD also assesses specifiers of HD diagnosis (insight and excessive acquisition). The first author CT was trained in the use of the SIHD and conducted the interviews. When the diagnosis was unclear, cases were discussed on an individual basis with author LFC, who was experienced in using the SIHD.

To establish HD severity, the *Hoarding Rating Scale Interview (*HRS-I, Tolin, Frost, & Steketee, 2010, Appendix 5.3) was rated by the carer. The HRS-I is a brief five-item measure which assesses the key features of HD (i.e. clutter, difficulty discarding, acquisition, distress and impairment) on a Likert scale from 0 (no problem/difficulty) to 8 (extreme problem/difficulty). A score of 14 or more indicates clinically significant hoarding symptomatology. The HRS has good reliability and validity (Tolin et al., 2010) and in the current sample the internal consistency was also good, α=.679. Additionally, carer's rated the *Clutter Image Rating scale (*CIR, Frost, Steketee, Tolin, & Renaud, 2008, Appendix 5.4). The CIR consists of a series of nine photographs of a kitchen, living room and bedroom with increasing levels of clutter in each photograph which the carers used to rate their relatives' house. A score of 4 or greater in any one room is usually taken as clinically significant levels of clutter and has good reliability and internal consistency (current sample α=.789) (Frost et al., 2008).

The *Family Impact Scale for Hoarding* (FISH, Nordsletten et al., 2014, Appendix 5.5) measures the specific impact of hoarding behaviours on the family including family accommodation to the hoarding (e.g. 'I avoid discarding my relative's possessions due to the distress it will cause him/her'). Participants are asked to rate statements on a 4-point Likert scale from 'strongly agree' to 'strongly disagree'. The FISH has been demonstrated to have good convergent and discriminant validity, and good internal consistency (current sample α=.829).

2.4.2. Self-report measures of carer experience

The *Experiences of Caregiving Inventory* (ECI, Szmukler et al., 1996, Appendix 5.6) is a 66 item measure used widely in the mental health caregiving literature to measure positive and negative experiences of caregiving (e.g. Addington, Coldham, Jones, Ko, & Addington, 2003; Murphy, Todd, & Joyce, 2001). The ECI consists of 10 subscales: two positive (rewarding personal experiences, good aspects of the relationship) and eight negative (negative symptoms, stigma, effects on the family, the need to provide back-up, dependency, problems with services, difficult behaviours, loss). Carers rate items on how often they have thought about the issue in the month prior to assessment on a scale from 0=never, 1=rarely, 2=sometimes, 3=often to 4=nearly always. The negative subscale has 52 items (maximum score=208) and the positive subscale is 14 items (maximum score=56) The ECI has good construct validity and reliability (Joyce, Leese, & Szmukler, 2000; Szmukler et al., 1996). In the current sample the internal consistency of the positive ECI was α=.866 and the negative ECI α=.958.

In order to assess the carers' appraisal of their relative, the relationship with the person with HD, and provide a measure of EE, the *Five Minute Speech Sample* (FMSS, Magaña et al., 1986; Malla, Kazarian, Barnes, & Cole, 1991) was coded. In comparison the to the 'gold standard' measure of EE – the Camberwell Family Interview (Leff & Vaughn, 1985) which takes between 1-2 hours to administer and another 2-3 hours to code (Hooley & Parker, 2006) – the FMSS provides a shorter alternative for identifying EE. In the FMSS, carers are asked to speak for 5 minutes about their thoughts and feelings about the person they care for. The speech is then coded for overall level of EE (high or low), criticism and emotional over involvement

(EOI). The FMSS has moderate reliability, although it may underestimate rates of high EE carers against the CFI (Hooley & Parker, 2006). The FMSS was rated between the first author (CT) and another researcher (EG). Both were trained on the FMSS by author JO. An interrater reliability analysis using the Kappa statistic was performed to determine consistency among raters which was found to be Kappa=0.75 (good reliability). Any differences on ratings were resolved through discussion and final ratings agreed.

Carer appraisals of the relative's hoarding behaviour were measured using the *Brief Illness Perception Questionnaire* (B-IPQ, Broadbent, Petrie, Main, & Weinman, 2006, Appendix 5.7) which assesses cognitive (e.g. expected timeline of illness) and emotional representations (e.g. level of concern) of an illness. Items 1, 2, 5, 6 & 8 represent negative illness representations and items 3, 4 and 7 are more positive beliefs about the illness (and are reverse scored for the total score). A modified version of the IPQ was found to be a reliable measure of models of illness in carers of patients with psychosis (Barrowclough et al., 2001). The B-IPQ has good test–retest reliability and concurrent validity (Broadbent et al., 2006). The internal consistency of the B-IPQ in the current sample was low α=.037.

Coping styles were assessed using the *Brief COPE* (Carver, 1997, Appendix 5.8). The Brief COPE consists of 14 subscales and carers rate how much they use each coping style with regard to a particular situation (in this case, the relative's hoarding). The Brief COPE has good internal reliability according to the original validation of the scale (Carver, 1997). Behavioural disengagement, self-distraction, substance use, and denial subscales were summed to represent an 'avoidant' coping style, in line with previous studies (Kuipers et al., 2006; Onwumere et al., 2011; Raune et al., 2004). In the current sample the internal consistency for avoidant coping was α=.656. The other two subscales considered to be less adaptive ways of coping are venting and self-blame. Active coping, emotional support, instrumental support, positive reframing, planning, humour, acceptance and religion subscales are considered to be more 'adaptive' coping strategies.

Levels of anxiety and depression in the carer (psychological distress) were measured with the *Hospital Anxiety and Depression Scale* (HADS, Zigmond & Snaith, 1983,

Appendix 5.9) which is a 14-item measure, seven of which assess anxiety symptoms (e.g. 'worrying thoughts go through my mind') and seven assess depression symptoms (e.g. 'I have lost interest in my appearance') over the last week. The anxiety and depression subscales can be analysed separately or combined to produce a measure of psychological distress. Zigmond and Snaith (1983) recommend a total score of ≥11 on either subscale as indicative of probable clinical levels of distress. The HADS has been used to assess psychological distress in carers of people with psychosis (e.g. Fortune, Smith, & Garvey, 2005) and it has well-established psychometric properties (e.g. Bjelland, Dahl, Haug & Neckelmann 2002). In the current sample the internal consistency was high for the total score (α=.910) and for anxiety and depression subscales (α=.858 and α=.877 respectively).

The psychosis comparison group had completed the General Health Questionnaire-28 item (GHQ-29; Goldberg & Hillier, 1979) which is also a brief measure assessing anxiety and depression. The 0, 0, 1, 1 method of scoring the GHQ-28 was used, with cut off of ≥5 indicating probable clinical levels of distress (Goldberg & Williams, 1988).

The *Warwick-Edinburgh Mental Well-being Scale (WEMWBS,* Tennant et al., 2007, Appendix 5.10) is a 14 item measure of mental wellbeing. Statements related to various aspects of wellbeing (e.g. 'I've been feeling relaxed') are rated over the last two weeks from 'none of the time' to 'all of the time'. The WEMWBS has good validity and reliability and has been demonstrated to have good responsivity to health interventions (Maheswaran, Weich, Powell, & Stewart-Brown, 2012). The internal consistency was high in the current sample (α=.928).

2.4.3. Group participant only measures

In addition to completing main outcome measures pre, post and at 1-month follow-up (with the exception of the SIHD, which was completed only at baseline), group participants completed a 'Knowledge about hoarding questionnaire' (developed by the authors) before and after the group. This was a 7-item multiple choice questionnaire about various aspects of HD including etiology and symptomatology (see Appendix 6).

After each group session, participants rated their satisfaction with the session on scale of 0-10 and gave overall feedback at the end of the group. In accordance with ethical approval, group facilitators actively checked in with participants regarding any difficulties arising from their attendance at the group. This system enabled one carer to seek additional support for himself and his relative with HD through facilitators' signposting him to local health and advice services.

2.5 Statistical Analyses

Statistical analyses were conducted in SPSS v22 for Windows (IBM Corp., 2013). For Phase 1, bivariate correlations were conducted to investigate the relationships between caregiver burden, distress, EE, coping and illness appraisals. The main outcomes of caregiver burden (measured by the ECI negative scale), positive experiences of caregiving (ECI positive scale) and distress (HADS or the GHQ-28) were compared with existing data on carers of people with psychosis (from Garety et al., 2008) using analysis of covariance in order to control for demographic variables. For this comparison, HADS and GHQ-28 scores were converted into z scores. Other variables will be descriptive and means and standard deviations will be reported.

For Phase 2, the pilot intervention, repeated measures analysis of variance were used to compare participant's pre, post, and 1-month follow-up outcomes on main variables of wellbeing (WEMWBS), distress (HADS), knowledge about hoarding, adaptive coping (Brief COPE), and appraisals of caregiving (ECI positive and negative scales). Additionally, paired t-tests between the different time-points (pre vs post, pre vs follow-up, and post vs follow-up) were also performed. Pre-post means and standard deviations of other variables and session by session measures will be reported.

2.6 Power calculations

A power calculation in G*Power (Faul, Erdfelder, Lang, & Buchner, 2007) showed that for within group comparisons of the four main factors in the caregiving model, a minimum sample size of 46 carers would have 80% power to detect a correlation coefficient of r=0.4 (medium effect size, two sided, alpha =0.05).

3. Results

3.1 Phase 1 Demographics

3.1.1. Demographics of HD carers

The majority of carers (n=53) were female (86.8%), with a mean age of 42.7 years (SD=13.64; range=20-77). All were UK-based, except one individual from the USA. In terms of ethnicity, broad categories showed that 81.1% were white and 18.9% from black or minority ethnic backgrounds. One person (1.9%) declined to state their ethnicity. These figures are representative of the population of the UK which was 86% white according to the 2011 census (Office for National Statistics, 2012). A more detailed breakdown of ethnicity is presented in *Table 1.*

Table 1: Ethnicity of the sample by categories used by the Office for National Statistics

Ethnicity	*n*	*%*
White British	41	77.4
White Other	2	3.8
Black British/Black African	2	3.8
Black British/Black Caribbean	2	3.8
Chinese	1	1.9
Pakistani	1	1.9
Mixed ethnic background	3	5.7

The educational history and employment status of participants are described in *Table 2*. Over half (56.6%) were working either full or part time, and a large proportion (86.8%) had completed higher education (above A-level).

Table 2: Educational history and employment status of the sample

Employment status	*n*	*%*
Employed full time	20	37.7
Employed part time	10	18.9
Voluntary employment	1	1.9
Unemployed	2	3.8
Housewife/husband	3	5.7
Student	6	11.3
Retired	5	9.4
Other (e.g. self-employed, sick leave)	6	11.3

Educational level (highest)	*N*	*%*
GCSE/16 years	2	3.8
A level	5	9.4
Undergraduate degree	19	35.8

Postgraduate degree	20	37.7
Other (e.g. diploma, BTEC)	7	13.2

Most participants were the adult children of parents with HD (n=35, 66.1%). A smaller proportion were partners or spouses (n=6, 11.3%), siblings (n=2, 3.8%), parents (n=2, 3.8%), and other relatives or friends (n=8, 15.1%). Less than one fifth of the sample (n=10, 18.9%) were living with the person with HD, but all were in regular contact; spending an average of 21.2 hours (SD=39.3; range=1-168) in weekly face-to-face or telephone contact with the relative. Participants had taken a mean 3.7 days off work in the last year to help their hoarding relative (SD=11.22; range=0-75). The current (2015) living wage in the UK is £7.85 an hour, and thus based on an average wage at this rate, 3.7 days would equate to £232.36 of lost earnings. However, 37 participants (69.8%) had not taken any days off work, so this finding was not applicable to all.

In terms of social support, the majority (n=42, 79.2%) reported that they had a confidant (someone they could talk to).

3.1.2. Carer-reported Hoarding Disorder demographics and characteristics

Table 3 describes the demographics of the relatives with likely HD. The average age of the person with HD was 65.9 years and over half were female. Carer estimates of the duration of their relative's hoarding problems varied between 5-70 years (median=30). Most carers described their relatives hoarding as being an issue 'for as long as I can remember'. The majority of relatives with HD were not in contact with mental health services.

The level of insight in hoarding difficulties was rated from the SIHD interview with the carer. 25 individuals with HD (47.2%) were rated as having good or fair insight into their problems, 18 (34%) had poor insight and 10 (18.9%) absent insight. The majority also acquired possessions excessively (n=49; 92.5%).

Table 3: Demographics of person with HD

	Mean	*SD*	*Range*
Age (years)	65.9	13.02	26-85
Estimated duration of hoarding in years*	29.4	15.28	5-70
	%	*N*	
Female	58.5	31	
Seen by mental health professional/team	11.3	6	

**Estimates by the carer could not always be specified; for example if parent had HD it may have begun before the carer was born or was very young.*

Table 4 describes the scores on the HD specific measures. On average the relative with HD scored well above the clinical cut-off on the HRS-I (Tolin, Frost, et al., 2010), and above the cut-off on the CIR (Frost et al., 2008) in all main rooms of the house. Average scores on the FISH were above the levels reported in the initial application of the scale (Nordsletten et al., 2014), indicating high levels of burden and family accommodation of symptoms.

Table 4: HD Characteristics

HD measure	*Mean*	*SD*	*Range*
HRS-I total	27.41	6.35	14-37
CIR Bedroom	5.96	2.04	1-9
CIR Living Room	5.55	2.07	1-9
CIR Kitchen	4.77	1.97	1-9
CIR Total	5.44	1.99	2.3-9
FISH Accommodation	14.42	3.37	10-22
FISH Burden	22.92	6.36	15-35
FISH Total	37.33	8.06	29-57

HRS-I = Hoarding Rating Scale-Interview, CIR=Clutter Image Rating scale, FISH=Family Impact Scale for Hoarding Disorder.

3.1.3. Demographics of psychosis carers comparison group (Garety et al., 2008)
Carers from the psychosis group (n=86) had a mean age of 52.91 years (SD=13.07; range=26-88) and most were female (n=59; 68.6%). The carers were predominantly white (n=73; 84.9%) and twelve (14%) were from black or minority ethnic backgrounds.

The largest group were parents of an adult service user with psychosis (n=43; 51.8%), followed by partners (n=29; 34.9%), with the remainder being siblings, children or other relations (n=11; 13.3%). Most carers lived with their relative (n=56; 65.1%). The average age of the relative with psychosis was 36 years (SD=11.93; range=18-64).

3.2 Phase 1 Results

All data were checked for normal distribution using Shapiro-Wilk tests and inspecting histograms and Q-Q plots. No variables violated the test of normality (Shapiro-Wilk p>.05).

Means and standard deviations of all measures for the HD carer group are reported in *Table 5.* The B-IPQ mean total was 56.96 (out of a possible 80) and the items relating to negative illness representations scored generally highly out of a possible 10. Participants gave personal control and treatment efficacy the lowest scores, indicating less optimistic beliefs about how much control they believed their relative had over their hoarding and that treatment would not be very effective. Participants reported a lower mean score on the WEMWBS than the average in the general population of 50.7 (Stewart-Brown & Janmohamed, 2008). *Table 6* displays the proportions of the sample classified as having high and low EE ratings.

Table 5: Means and Standard Deviations of all variables

Variable	*Mean*	*SD*
ECI positive	21.26	9.41
ECI negative	86.83	36.97
B-IPQ total	56.96	6.48
1.IPQ Consequences	7.91	1.66
2. IPQ Timeline	9.23	1.59
3. IPQ Personal control^{R}	2.68	2.03
4. IPQ Treatment efficacy^{R}	3.58	2.89
5. IPQ Identity	5.83	2.64
6. IPQ Concern	8.38	1.76
7. IPQ Understanding^{R}	5.81	2.95
8. IPQ Emotional Response	7.70	2.02
Brief COPE avoidant	13.25	3.64
COPE Distraction	4.32	1.87
COPE Denial	2.53	1.05
COPE Substance Misuse	2.42	1.10

COPE Disengagement	3.98	1.66
COPE Venting	4.08	1.40
COPE Self Blame	3.53	1.78
HADS total	14.87	8.56
WEMWBS	29.81	9.49

ECI= Experiences of Caregiving Inventory, IPQ=Brief Illness Perceptions Questionnaire, HADS=Hospital Anxiety and Depression Scale, WEMWBS=Warwick-Edinburgh Mental Wellbeing Scale. ^{R}*=Items reverse scored for total IPQ.*

Table 6: Classifications of Expressed Emotion in the sample

Expressed Emotion Rating	*N*	*% of sample*
Low	24	45.3
High	29	54.7
High : Critical	22	41.5
High : EOI*	4	7.5
High : Both critical and EOI	3	5.7

**EOI=Emotional over involvement.*

3.2.1. Carer distress, illness appraisals and caregiving burden (Hypothesis 1)

As *Table 7* shows, Pearson's correlations showed large significant positive associations between distress, appraisals and burden. Thus, the more negative illness appraisals, the greater the level of burden and distress, and vice versa.

Table 7: Correlations between appraisals, caregiver burden and distress

Variable	*Appraisals (IPQ)*	*Burden (ECI Negative)*	*Distress (HADS)*
Appraisals	1	-	-
Burden	.572**	1	-
Distress	.572**	.574**	1

***P<.001*

3.2.2. Carer distress, burden, relationship quality and coping (Hypothesis 2)

A Pearson's point-biserial correlation showed that high EE was associated with higher levels of carer distress, but not with avoidant coping[4] (r_{pb} =.157, *p*=0.26). Distress was also not significantly associated with avoidant coping. Results are presented in *Table 8*. Further analyses of individual brief-COPE subscales indicated that Venting coping styles were found to be significantly associated with EE (r_{pb} (51)=.351, *p*=0.01). The remainder of the less adaptive individual subscales were not significantly associated with EE (r's range=-.03 to .24).

Table 8: Associations between EE, avoidant coping and distress

Variable	*Relationship quality (EE)*	*Avoidant coping*	*Distress (HADS)*
Relationship quality	-		
Avoidant coping	.157 (p=.263)	-	-
Distress	.308*	.233 (p=.093)	-

**p<.05*

An independent-samples t-test showed that high EE critical carers (but not high EE overall when EOI was included) reported significantly higher levels of caregiver burden (mean=101.64; SD=36.30) than low EE carers (mean=76.8; SD=36.86) (t(44) -2.3, *p*=0.026, 95% CI [-46.61, -3.08]).

3.2.3. Comparisons of levels of distress and caregiving experiences between participants and psychosis carers (Hypothesis 3)

Independent samples t-test and Chi-square analyses were conducted to compare the psychosis and HD carers on demographic variables of age, gender and ethnicity. HD carers were significantly younger (p<.001) and had significantly more females (p=.02). Analyses were thus conducted with gender and age as covariates.

[4] For avoidant coping: High EE mean=13.75, Low EE mean=12.62.

An analysis of covariance (ANCOVA) showed that there was no significant difference in scores on the negative scale of the ECI (caregiver burden) between psychosis carers and HD carers after controlling for age and gender, indicating similar levels of burden in the two groups (see *Table 9*). However, HD carers reported significantly less positive experiences of caring and appraisals of the relationship (ECI positive) than psychosis carers.

Twenty-three (out of 53; 43.4%) of HD carers reported clinical levels of distress (41% above cut-off for anxiety, 9.4% for depression), compared to 55.1% (n=38/69) of psychosis carers. An ANCOVA with the converted z scores showed that there was no significant difference between the groups on distress (*Table 9*).

Table 9: ANCOVA comparison between HD and psychosis carers on the ECI

	HD carers (n=53)	Psychosis carers (n=63)		
	Mean (SD)	Mean (SD)	F (df)	*P*
ECI negative	86.8 (36.97)	94.6 (31.43)	.648 (1, 112)	0.423
ECI positive	21.26 (9.41)	31.15 (7.86) (n=69)	26.79 (1,104)	<.001
Distress (HADS/ GHQ-28)	-	-	.007 (1,108)	.934

3.2.4. Summary of Phase 1 results

In summary, hypotheses were mostly supported. Negative appraisals of the relative's illness and behaviour were significantly associated with both higher levels of carer burden and higher levels of distress. EE was significantly associated with distress but not avoidant coping. Carers of people with HD had similar levels of distress and caregiving burden to carers of people with psychosis. Indeed, HD carers actually had significantly less positive experiences of caregiving than psychosis carers.

3.3 Phase 2 Descriptives

3.3.1. Demographics of carers in the psychoeducational group

Twelve participants from Phase 1 also attended the group intervention. The mean age of participants was 40.17 years (SD=16.13, range: 21-73) and three-quarters (75%) were female. Eight participants (66.7%) were white British, and four (33.3%)

were from black and minority ethnic groups. Half of the group were married or cohabiting (n=6) and the remainder were split evenly between those who were single (n=3) and separated or divorced (n=3). Half of the sample were working full-time or part time (n=6) and the rest were either students (n=2), unemployed (n=2), volunteering (n=1) or retired (n=1).

Nine participants (75%) were the children of a parent with HD, one (8.3%) participant was a parent of a son with HD, one (8.3%) was a nephew of an uncle with HD and for one participant the relative with HD was their ex-stepmother (8.3%). Two members of the group were currently living with their hoarding relative (16.7%). The mean number of hours of contact with the relative was 15.25 a week (SD=15.78; range=1-50).

3.3.2. Sessions attended and data completion

The mean number of sessions attended by participants was 4.92 (SD=0.99; range=3-6) out of a total 6. Full data sets were completed for eight participants (i.e. baseline, post and 1 month follow up). One participant did not complete post-group measures and three did not complete the 1-month follow-up measures.

Demographics and HRS-I total scores of participants who provided full data sets versus those who missed 1 or more data points are detailed in *Table 10*. Mann-Whitney U and chi-square goodness of fit comparisons between the groups revealed that people who did not complete all measures were more likely to be white British and not living with their hoarding relative. However, given the small sample sizes of the compared groups, these differences in demographics are likely to be negligible. Importantly, there were no differences in the severity of the hoarding symptoms of their index relative as measured by the HRS-I (p=0.72).

Table 10: Comparison of participants with and without full data sets

	N (%) or M (SD)		*p value*
Variable	*Participants with full data sets (n=8)*	*Participants with any missing data (n=4)*	
Gender (Female)	6 (75%)	3 (75%)	.083
Age	36.63 (16.71)	47.25 (14.22)	.283
Ethnicity			.050*

White British	5 (62.5%)	3 (75%)	
Black Caribbean	2 (25%)	0	
Mixed/multiple ethnic background	1 (12.5%)	1 (25%)	
Employed full or part time	3 (37.5%)	3 (75%)	.306
Married or cohabiting	4 (50%)	2 (50%)	1
Living with HD relative	2 (25%)	0	.021*
HRS-I total score	25.38 (6.72)	31.50 (3)	.073

**significant at p<0.05 level*

3.4 Phase 2 Results

To investigate the impact of the intervention on carer wellbeing, distress, coping, and caregiving experiences, repeated measures analysis of variance (ANOVA) tests (sphericity assumed) were conducted, with pairwise comparisons using the Bonferroni correction, to investigate effects between pre, post and follow-up. Again all data were normally distributed. The eight participants who completed all measures were included in the repeated-measures ANOVA analysis. Paired t-tests were used for the knowledge about hoarding questionnaire, as this was administered only twice (before and after the group). Means, standard deviations and *p* values for the ANOVA are presented in *Table 11.* Paired t-tests were also conducted for a fuller data set (n=11 at pre-post), the results of which are presented in Appendix 7.

3.4.1. Effects of the group on understanding of HD (Hypothesis 1)

A paired t-test was carried out to compare knowledge about hoarding before and after the intervention (n=11).

There was a significant increase in the scores on the knowledge about hoarding questionnaire from pre-intervention (mean=4.81, SD=0.98) to post intervention (mean=5.81, SD=0.60) (t(10)=-2.08, p = 0.02) for the eleven carers who completed the questionnaire both before and after the intervention.

3.4.2. The effect of the group on reports of carer wellbeing, distress and aspects of caregiving (Hypothesis 2)

A) Wellbeing

The results from the repeated measures ANOVA (*Table 11*) showed that mean wellbeing differed significantly between time points ($F(2,14) = 16.62$, $p<0.001$). Pairwise comparisons revealed a non-significant improvement from pre to post intervention, but a significant improvement between pre-intervention and 1-month follow-up and between post-intervention and 1-month follow-up. The t-test for the fuller sample showed that with eleven participants there was also a significant improvement from pre to post (Appendix 7).

B) Distress

There was no significant change in distress levels over the 3 time points on the ANOVA ($F(2,14)=.212$, $p=0.81$) indicating the intervention had no effect on levels of distress. The same results was obtained with paired t-tests.

C) Caregiving experiences

Positive experiences of caregiving (both positive appraisals of the relationship and positive personal experiences) differed significantly between time points ($F(2, 14)=9.18$, $p=0.003$). Pairwise comparisons revealed that positive aspects of caregiving improved significantly from pre to post intervention, an improvement which was maintained at 1 month follow-up. The same results was obtained with paired t-tests.

There was no significant change in caregiver burden following the intervention ($F(2,14)= 0.629$, $p=0.547$).

3.4.3. Coping styles (Hypothesis 3)

There were no significant differences in use of either avoidant or adaptive coping strategies after the intervention (p's>.05). The paired t-tests showed a significant result at post to follow-up for emotional support coping style ($t(7)=-3.00$, $p=0.02$).

Table 11: Means and standard deviations, ANOVA results and pairwise comparisons for group outcomes (N=8)

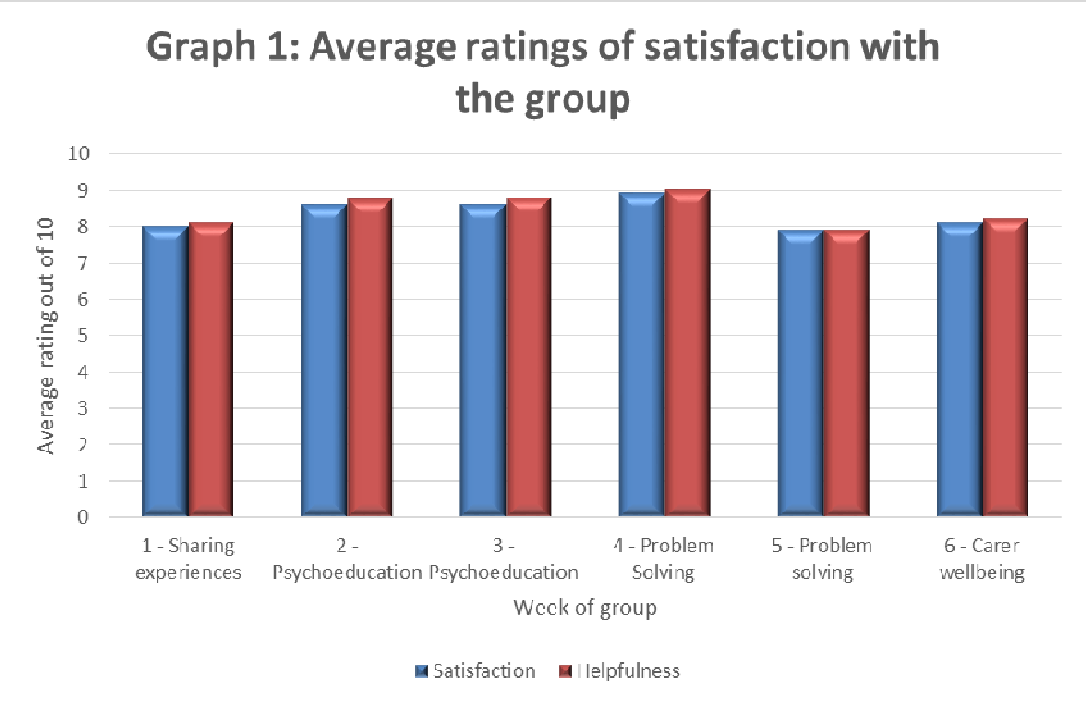
Variable	*Mean (SD)*				*Pairwise comparisons: mean difference (p value)*		
	Pre-intervention	*Post-intervention*	*1-m FU*	*p=*	*Pre-post*	*Pre-FU*	*Post-FU*
Wellbeing (WEMWBS)	29.5 (8.12)	33.63 (11.64)	42.25 (8.92)	<.001*	-4.125 (0.229)	-15.75 (0.004)*	-11.625 (0.026)*
Distress (HADS)	12.13 (5.77)	12.63 (6)	13.25 (4.27)	.811		n/a	
ECI negative	79.75 (22.52)	83.37 (28.34)	89.00 (30.36)	.547		n/a	
ECI positive	20.50 (8.45)	26.13 (10.43)	25.63 (9.27)	.003*	-5.625 (0.015)*	-5.125 (0.017)*	.500 (1)
Avoidant coping (brief-COPE)	12.75 (2.71)	12.63 (3.50)	12.50 (2.27)	.979		n/a	
Active coping	5.13 (2.17)	5.88 (1.25)	5.25 (1.67)	.481		n/a	
Emotional support	4.63 (1.77)	4.50 (1.31)	5.25 (1.49)	.123		n/a	
Instrumental coping	4.75 (1.83)	5.00 (2)	4.75 (1.83)	.873		n/a	
Reframing	3.25 (0.89)	3.50 (1.07)	3.38 (1.50)	.847		n/a	
Planning	5.38 (1.06)	6.00 (1.07)	5.38 (1.30)	.463		n/a	
Humour	3.75 (1.28)	3.25 (1.28)	3.50 (1.31)	.595		n/a	
Acceptance	4.75 (1.49)	4.88 (1.46)	5.13 (1.36)	.706		n/a	
Religion	3.00 (1.51)	3.25 (1.39)	3.50 (1.60)	.321		n/a	

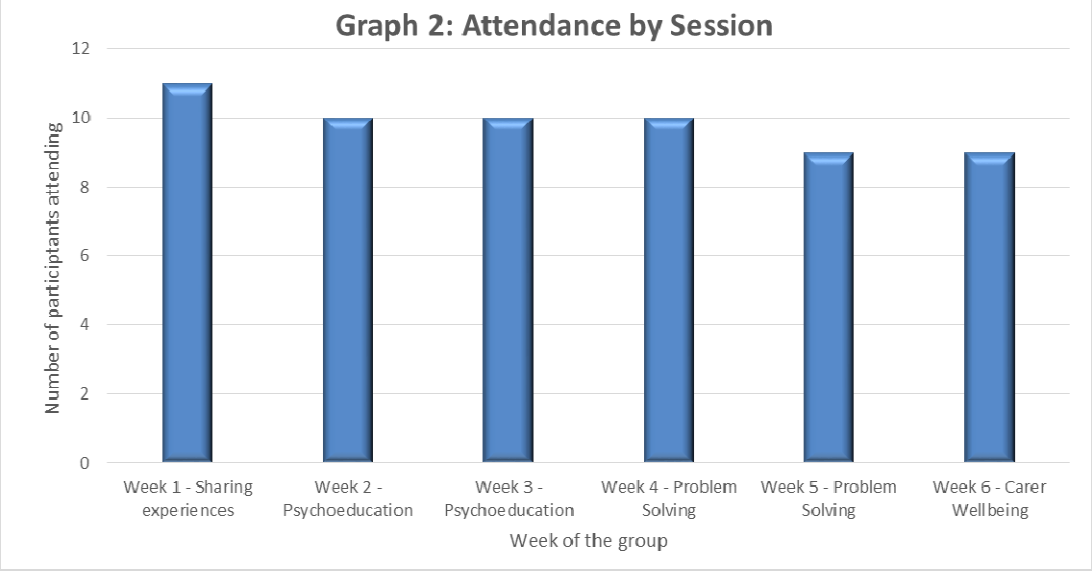
**significant at p<0.05 level*

3.4.4. Weekly satisfaction rating (Hypothesis 4)

Graph 1 shows average ratings of satisfaction and perceived helpfulness by week of the group, rated on a scale of 0-10. Overall satisfaction (mean=8.35; SD=0.40) and

helpfulness (mean=8.46; SD=0.45) ratings were high over the duration of the group. Brief qualitative feedback was elicited from group participants at the end of the intervention. Participants reported a range of gains, including the benefit of meeting others in a similar position "*Just knowing that other people have felt similar things to me has made me feel so much better about the situation*", the usefulness of the psychoeducational material "*I found out many facts about hoarding which I did not know at all*", and the practical problem solving sessions "*I will definitely go into active sorting through stuff with my dad with more patience and more understanding and empathy*". Suggestions made by participants of how to improve the group included: wanting additional information on working with professionals, more practical *"techniques for dealing with and making progress with helping a hoarder"* and some people wanted a greater number of sessions. Participants suggested that the group size was suitable, but reported that when some people missed a week it felt too small. Attendance by week of the group is presented in Graph 2.





3.3.5. Summary of Phase 2 results

Carer reported understanding of HD improved after the intervention. Wellbeing and positive experiences of caregiving also improved after the intervention and gains were maintained at 1-month follow-up. There was no change in distress, burden or coping styles after the group in the main analysis.

4. Discussion

This study aimed to investigate the experiences of carers of people with HD and examine the feasibility and effectiveness of a brief CBT-based psychoeducational group intervention. It was hypothesised that carers of people with HD would have high levels of distress and caregiving burden, at least equivalent to carers of people with psychosis, that negative appraisals of HD would be associated with carer distress and burden, and that EE would be associated with poorer coping and distress. The group intervention aimed to improve carer's knowledge about HD, wellbeing, distress, coping and experience of caregiving.

4.1 Phase 1

Over half of carers who participated in the study were female adult children of people with HD, although there was also a fair proportion of other relationships such as partners/spouses. This is roughly in line with previous studies in HD (Drury et al., 2014; Tolin, Frost, Steketee, & Fitch, 2008). This caregiving population is somewhat different from the comparison group of psychosis carers who were mostly parents or partners of the individual with psychosis and more often living with the person.

There is a paucity of research into the particular needs of adult children of people with mental health problems. However, we do know that they can experience elevated rates of psychological problems (e.g. depression) and functional difficulties (Bijl, Cuijpers, & Smit, 2002; Mowbray, Bybee, Oyserman, MacFarlane, & Bowersox, 2006; Pilowsky, Wickramaratne, Nomura, & Weissman, 2006). In HD the picture may be similar; Drury et al.'s (2014) family members reported functional impairment equivalent to that of their hoarding relatives. The current study recorded high levels of psychological distress and burden for adult children.

Key issues for children of people with HD are often related to the secrecy, embarrassment and stigma around the hoarding, concerns about their relatives' safety and fears of becoming a 'hoarder' themselves (Park et al., 2014; Sampson, 2013; Wilbram et al., 2008). Park and colleagues (2014) suggested that some children of people with HD may even terminate contact with the relative as a result of many years of frustrated efforts to change their relative's hoarding behaviours. The current study excluded participants without current contact with the relative, though this would be an interesting area for future research.

To the authors' knowledge, this is the first study to date to investigate different aspects of the caregiving role in HD. The hypotheses were mainly supported; carers held threatening appraisals of the illness and few positive appraisals, which were related to high levels of burden and psychological distress. Over half of all carers had high EE, which was also associated with greater levels of distress. Before this study, there was very little research on caregiver burden in HD, with the exception of Drury et al. (2014) who identified levels of caregiver burden in HD equivalent to or above those reported by carers of people with Alzheimer's disease. The current study adds to the literature on family experiences by investigating theoretically based sources of burden and distress in carers, including the role of expressed emotion and illness appraisals, both of which have been studied extensively in other disorders (Kuipers et al., 2010).

The present study identified high levels of affective disturbance and burden in HD carers. These were contextualised by comparing results to psychosis carers, in whom caregiver experience has been widely documented, and where similarities between

the two conditions can be observed. The findings revealed levels of anxiety and depression falling within the clinical range and caregiving burden similar to that of carers in the psychosis comparison group. Furthermore, carers had significantly less positive experiences of caregiving, such as satisfaction and personal gains from the role, than psychosis carers and poorer wellbeing than the general population (Stewart-Brown & Janmohamed, 2008). Results from the measure of impact of HD on the family (FISH; Nordsletten et al., 2014) indicated a substantial level of carers' accommodation of HD symptoms and HD specific burden, even above the levels reported in Drury et al. (2014). Participants reported a number of ways in which hoarding affected them including negative impact on their social life, feeling upset and frustrated by the hoarding, feelings of embarrassment and guilt, conflict in the relationship including verbal aggression and concern about the relatives' health.

The potential presence of high EE in HD carer relationships was indicated by previous research which showed high levels of patient rejection (Tolin, Frost, Steketee, & Fitch, 2008) and conflict between carers and their hoarding relatives (Park et al., 2014). This study found that over half of carer relationships were classed as high EE, mainly critical, which represents similar levels to those found in OCD carers (e.g. Chambless & Steketee, 1999) and psychosis carers (e.g. Cechnicki et al., 2013). The associations between high EE, burden and distress are in line with previous research into carers of people with psychosis (Kuipers et al., 2006; Raune, Kuipers, & Bebbington, 2004; Scazufca & Kuipers, 1996). Only critical high EE carers had higher levels of burden than low EE; emotional over-involvement was not associated with higher burden. However, this may be an artefact of the small size of this group of carers, as only four participants were classified as EOI.

A key factor in understanding the reports of caregiver burden and distress is the role of illness appraisals. In the present study HD carers held many threatening appraisals of HD (e.g., that the consequences of the disorder would be severe, that it would last a long time, that their relative had little control over it). Negative illness appraisals were associated with higher burden and distress, consistent with findings in other severe mental health conditions (Addington et al., 2003; Barrowclough, Gooding,

Hartley, Lee, & Lobban, 2014; Barrowclough et al., 2001; Fortune et al., 2005; Jansen et al., 2015; Onwumere et al., 2008).

In contrast with much of the literature in severe mental health conditions (e.g. Fortune et al., 2005; Kuipers et al., 2006; Onwumere et al., 2011; Perlick et al., 2008), we did not find that avoidant coping was related to either increased distress or EE, with the exception of venting styles of coping, which were associated with high EE. Venting could be argued to be conceptually similar to EE in terms of focus on the negative aspects of the relationship. It seems plausible therefore that relatives who tended to employ a venting coping style might have reported a greater number of critical comments within the FMSS (Magaña et al., 1986). Previous qualitative research in HD had indicated that family members may use avoidant coping strategies (Wilbram et al., 2008). However, in this study we did not find particularly high levels of avoidant coping; indeed mean levels of avoidant coping were slightly lower than has been previously reported in psychosis carer groups (Onwumere et al., 2011; Raune et al., 2004). Perhaps the fact that individuals signed up to the research could be a soft indicator that this group were not avoidant copers. Further research into the coping styles used by HD carers would be of benefit.

Overall, the findings are mostly in keeping with the cognitive model of caregiving (Kuipers et al., 2010), with some notable differences. In accordance with the model, negative appraisals of the illness seemed to be positively linked to more negative appraisals of caregiving (i.e. burden) and greater distress. In turn high EE (an approximation of current relationship quality) was prevalent and also associated with distress and burden. However, there was no evidence for a differential impact on coping strategies in the current study. Another difference to the model is that due to the nature of HD often being a chronic disorder, which typically begins in adolescence (e.g. Grisham, Frost, Steketee, Kim, & Hood, 2006), HD is likely to have been a longstanding presence in the relationship between the patient and carer (particularly for children of parents with HD). Therefore, the influence of the 'previous relationship' pre onset of HD may be less relevant here. It is possible that the lack of positive caregiving experiences and high levels of burden may reflect the

relative absence of available support and information for families and that many relatives with HD had low insight and were mainly not help seeking.

4.2 Phase 2

The results of the pilot six-session group intervention provide further preliminary evidence that carer-only interventions have merit and could be of value in HD. Group participants were mainly female adult children of a parent with HD and drawn from a range of ages and ethnicities. Only two participants were currently living with the hoarding relative. As with previous studies, there was some dropout in completing measures, particularly at follow-up, although attendance at group sessions was good. Nevertheless, this was the largest study to date to evaluate a carer-only intervention in HD. There were some statistically significant differences between those who completed and did not complete the intervention. This finding should be interpreted with caution, given the very small sample size, although it is possible that people living with their hoarding relative were more motivated to participate.

Hypotheses were partially supported; following group attendance, there were significant improvements in reports of positive caregiving experiences, wellbeing and understanding of hoarding in carers. Levels of avoidant coping strategies, distress and caregiving burden remained unchanged. Quantitative and qualitative feedback suggested that the group was well-received by participants and that the content and length of the group were acceptable.

The improvements in wellbeing are particularly encouraging, given that baseline levels fell below average (Stewart-Brown & Janmohamed, 2008). Research in other disorders (namely schizophrenia spectrum conditions) has found that family wellbeing can be increased by involving carers in interventions (e.g. Berglund, Vahlne, & Edman, 2003; McFarlane, Dixon, Lukens, & Lucksted, 2003). However, it is more unusual for a brief intervention, like the present study, to have a positive impact on wellbeing (Lucksted, McFarlane, Downing, & Dixon, 2012). One possible mechanism for increased wellbeing may have been the group setting. The group was designed to be a safe and supportive environment where participants were able to share their experiences of having a relative with HD. Mutual support from others

with similar experiences, with whom they could share ideas for coping and managing the problem, and recognise that they were not alone in their concerns and frustrations, may have helped to enhance wellbeing (Chien & Norman, 2009; Citron, Solomon, & Draine, 1999; Cuijpers, Hosman, & Munnichs, 1996). Furthermore, the problem solving sessions (weeks 4 & 5) and the final session on promoting carer wellbeing (week 6), were designed to help carers recognise their own needs and the importance of maintaining their own wellbeing in order to provide support for their relatives.

Another important finding was the increase in positive experiences of caregiving, from a baseline level significantly lower than that of the psychosis comparison group (Garety et al., 2008). There has been a fairly recent move in the caregiving literature to acknowledge the presence and importance of positive caregiving experiences (also described as 'caregiver rewards' or 'caregiver satisfaction') as well as burden, and to focus on building strengths as well as reducing problematic aspects in interventions (e.g. Bauer, Sterzinger, Koepke, & Spiessl, 2013; Onwumere et al., 2008; Veltman, Cameron, & Stewart, 2002). Research has shown that positive experiences of caregiving were associated with lowered depression and burden, better reported health (Cohen, Colantonio, & Vernich, 2002), and better quality of life (Kate, Grover, Kulhara, & Nehra, 2013). Therefore, improvements in positive appraisals of caregiving were potentially related to increased wellbeing.

Means by which positive experiences of caregiving improved may have been through increased social support, improved interactions with their relative and a better understanding of HD. In a large sample of carers, Chen and Greenberg (2004) found that formal support from professionals and informal support from participation in support groups were positively associated with caregiving rewards. Relatives of people with HD are particularly unlikely to receive professional support due to the dearth of services for HD, and even less available support for families. Members of the group reported having very few people whom they could talk to about their relatives hoarding and, as previously discussed, HD is associated with stigma and embarrassment which can add to the isolation of relatives. Participants in Sampson et al.'s (2012) group also identified the social support element as helpful.

Anecdotally, group participants reported that they were more able to have more constructive conversations with their relatives and also felt more confident in speaking to others about HD.

Improved understanding of HD may have been a route through which positive aspects of caregiving and/or wellbeing improved. One aspect of the ECI is related to understanding the relatives' mental health condition and feeling more empowered to help the person as a result. The psychoeducational nature of the group enabled learning about the symptoms of HD, the proposed causes and treatments. Group members reported finding this information invaluable, particularly given that there was little information available on the disorder. This finding along with evidence from the previous HD carer intervention studies (Chasson et al., 2014; Sampson et al., 2012) indicates that an important aim of interventions should include developing a better understanding of HD.

No improvements in distress or carer burden levels were observed following the intervention, contrary to predictions. Several researchers have noted that psychoeducational carer interventions reliably improve knowledge, coping and sense of social support, but can struggle to reduce burden and distress levels (see review by Sin & Norman, 2013). Szmukler and colleagues (2003) suggested that burden and psychological distress may only improve once the relatives' symptoms reduce. The lack of change in coping strategies, aside from a change from post to follow-up in emotional coping, is in contrast to the findings of Chasson et al. (2014) who observed an increase in some adaptive coping strategies and reduction in less helpful strategies after their intervention for carers of relatives with HD. The small improvement in emotional coping is an encouraging finding and merits further study.

Participants indicated that the inclusion of psychoeducation, a supportive group setting and 'practical' problem solving tips were important ingredients of the group. As was suggested by Sampson et al. (2012), carers in the current group indicated that a group size of between 6 and 8 is preferential to allow sufficient time to talk, but not be too small. Feedback also indicated that professional facilitation was helpful. Some participants indicated a wish for more emphasis on practical strategies

to help their hoarding relative e.g. how to support them to declutter. A longer term follow-up to determine whether changes were maintained, or additional 'booster' sessions may be useful for future interventions.

4.3 Strengths

The current studies had a number of strengths. First, the direct recruitment of carers via publicly accessible forums rather than through professional services meant that our sample was likely to be less biased toward carers whose relatives with HD had better insight. Equally, we may have recruited a more highly distressed and help seeking section of relatives. Second, the study utilised widely-used validated measures with good psychometric properties, which allowed comparison with other groups of carers. Third, the comparison with psychosis also helped to set the findings in context; too often research focuses on a single diagnosis which limits interpretation. Fourth, the group intervention used a theoretically informed protocol which targeted several of the components identified in a recent systematic review of carer interventions to be most helpful for carers of individuals with severe mental health conditions (Lobban et al., 2013), including stress management, psychoeducation, problem solving and social support.

4.4 Limitations

A limitation of Phase 1 was the cross-sectional design. In the absence of a longitudinal study, conclusions about causality or direction of effect remain unknown. Another important limitation was that we did not interview the hoarding relative directly, and thus cannot be 100% sure that they met DSM-5 criteria for HD. It is also possible that carer's reports of clutter and HD symptoms may not have been accurate, as we did not require photographs of the relative's home, and previous studies have found that family and friends rate may symptoms as more severe than the individual with HD (e.g. Dimauro et al., 2013).

A couple of limitations relate to measurement protocol; for example, the SIHD and FMSS were conducted via telephone. The absence of the face to face assessment may have impacted on the amount that participants reported; for example, participants may have reported more about their relationship on the phone. That said, previous studies have found good correspondence between telephone and

face-to-face administration of the FMSS (Beck, Daley, Hastings & Stevenson, 2004). To ensure a broader participant sample where geographic distance in relation to the educational institution undertaking the study is minimised, further studies exploring the outcomes of telephone based interview assessment in relation to face to face meetings would be helpful. The second limitation of measurement was the low alpha value of the B-IPQ total score. This is partially to be expected, given that in the brief version of the IPQ, each question represents a separate scale (Broadbent et al., 2006), but further validation of this questionnaire is warranted in this population. Future research could examine which particular negative appraisals are associated with higher distress levels.

With regard to the sample in phase 1, there was large variability in the amount of contact between the carer and the individual with HD (range=1-168 hours a week). Over 10 hours of caregiving per week has been associated with a decline in mental health in carers (Smith et al., 2014) and it is possible that carers with greater levels of contact would have been affected differently by the hoarding. Future studies may usefully investigate whether amount of contact makes a difference to the experience of caregivers in HD.

Though novel, the small sample size of the pilot intervention (phase 2) and uncontrolled design mean that the positive results require replication in a larger scale study. There was some attrition in completing measures at post and follow-up, although there was less dropout than in previous intervention studies (Chasson et al., 2014; Sampson et al., 2012). Given that there is very little support and information available for relatives of people with HD in the UK, it could be argued that the positive outcomes may have been due to non-specific effects and the peer support element of the group. A final limitation of phase 2 was that the 'Knowledge about Hoarding' questionnaire was not a validated measure.

4.5 Future research

4.5.1. Phase 1

Further research is indicated to attempt to understand the coping strategies used by carers of people with HD (including the use of social support) and how these might be linked to carer burden and distress. This study has provided further evidence that

family relationships can be disrupted, and future research could usefully investigate both the problematic and positive aspects of relationships. Medard and Kellett (2014) identified elevated levels of attachment anxiety and unusual attachment to objects in people with HD. Insecure attachment styles in people with HD are likely to influence interactions with family members, and it is established that both insecure parental attachment style and parental anxiety disorders confer a risk of insecure attachments in their children (Manassis, Bradley, Goldberg, Hood, & Swinson, 1994; van IJzendoorn, 1995). Attachment and interpersonal styles are worthy of future study in both people with HD and their relatives. Related to this, it would be useful to identify the prevalence of children and adolescents growing up with a parent with HD and the impact of this on their social and emotional development. Recently there has been increasing recognition of the needs of 'young carers' (children under 18) and the importance of providing support for this group in other disorders (e.g. Cooklin, 2006, 2010). Further study is warranted on 'young carers' in HD.

4.5.2. Phase 2

The next step for future research in terms of carer interventions in HD would be to replicate the group intervention with a larger sample and develop the manual. If results continue to prove promising, a randomised control design with a control group would be indicated to help to determine the efficacy of the intervention. Examination of the mechanisms of positive change and key components would be indicated. Results from phase 1 could inform improvements to future interventions. For example, future intervention studies may target the relationships between carer and individual with HD, as we identified a high rate of EE and particularly critical relationships. Future interventions could also include components designed to decrease levels of anxiety and depression in carers and target carer wellbeing and positive appraisals of caregiving, as these were identified to be particularly low at baseline in the whole group of carers.

Future studies could usefully investigate whether carer interventions have an effect on coping strategies, as findings have been inconsistent thus far. Furthermore, researchers could investigate whether carer interventions have any direct or indirect effect on HD symptoms in the relative. Chasson et al. (2014) reported on carer-rated

HRS scores pre and post, although no changes were observed. Another future step would be to develop and pilot family interventions involving both the person with HD and their relatives, which have proved successful in a number of other disorders (Pharoah et al., 2010; Pinsof & Wynne, 1995) and could possibly be useful in improving outcomes for HD (e.g., improving engagement and motivation). Family and carer-only interventions also have the potential to improve relationships and communication; future studies should measure these outcomes.

4.6 Clinical Implications

4.6.1. Phase 1

Although the evidence base is still in the early stages regarding our understanding of the needs of HD carers, the current study has further demonstrated the profound impact of HD on relatives. As understanding of HD and its effects continues to progress and specialist services are developed, the needs of families should be routinely taken into account. In other disorders, families are viewed as instrumental for recovery and in need of support. For example, in psychosis, family interventions and carer support are recommended in the NICE treatment guidelines in England and Wales and more globally (Gaebel, Weinmann, Sartorius, Rutz, & McIntyre, 2005; NICE, 2014). This study has identified a clinical need for psychological support, or at the very least provision of good, evidence based information, tailored to the needs of relatives of people with HD in order to improve understanding of the condition and address common concerns. Knowledge of HD is not widespread in the general public or even amongst professionals. Thus, researchers should seek to increase public knowledge of the condition and agencies should work together to create joint hoarding 'task forces' (Bratiotis, 2013) to tackle the environmental, physical and, psychological effects of HD. Professionals should be aware that there may be children living in a home with substantial clutter which can pose a risk to their physical, social, and emotional wellbeing during their younger and adult years.

4.6.2. Phase 2

The results of the group intervention indicate that a brief intervention for carers maybe have important effects in improving knowledge, wellbeing and positive experiences as part of their caregiving role. Interventions should be tailored to support carers with the specific challenges posed by having a relative with HD. The

present study indicated that positive relationships can and do exist between people with HD and their families, and that carers have positive ways of coping with the situation which interventions should aim to build on. Members of the current group and in Sampson et al.'s (2012) study indicated that ongoing support for family members is desired in addition to brief focussed interventions.

4.7 Conclusion

In summary, caregivers of people with HD experience high levels of stress and burden, equivalent to those found in psychotic conditions, which are themselves recognised as globally burdensome (Whiteford et al., 2013). The burden and distress reported by HD carers were associated with more negative appraisals of their relatives hoarding. Many carers had relationships characterised by high expressed emotion which added to distress and burden. The results of the pilot intervention indicate that a brief psychoeducational group may improve the experiences and wellbeing of carers, which can be maintained at one-month after the group. The study requires replication in a larger controlled trial. However, current findings from Phase 1 and 2 of this study reiterate the need for recognition of the impact of HD on families and provision of family support.

5. Bibliography

Addington, J., Coldham, E. L., Jones, B., Ko, T., & Addington, D. (2003). The first episode of psychosis: the experience of relatives. *Acta Psychiatrica Scandinavica*, *108*(4), 285–289. doi:10.1034/j.1600-0447.2003.00153.x

Ale, C. M., Arnold, E. B., Whiteside, S. P. H., & Storch, E. A. (2013). Family-Based Behavioral Treatment of Pediatric Compulsive Hoarding: A Case Example. *Clinical Case Studies*, *13*(1), 9–21. doi:10.1177/1534650113504487

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders: DSM-5.* (5th ed.). Washington, D.C.: American Psychiatric Association.

Amir, N., Freshman, M., & Foa, E. B. (2000). Family distress and involvement in relatives of obsessive-compulsive disorder patients. *Journal of Anxiety Disorders*, *14*(3), 209–217. doi:10.1016/S0887-6185(99)00032-8

Awad, A. G., & Voruganti, L. N. P. (2008). The burden of schizophrenia on caregivers: a review. *PharmacoEconomics*, *26*(2), 149–162. doi:10.2165/00019053-200826020-00005

Barrowclough, C., Gooding, P., Hartley, S., Lee, G., & Lobban, F. (2014). Factors associated with distress in relatives of a family member experiencing recent-onset psychosis. *The Journal of Nervous and Mental Disease*, *202*(1), 40–6. doi:10.1097/NMD.0000000000000072

Barrowclough, C., Lobban, F., Hatton, C., & Quinn, J. (2001). An investigation of models of illness in carers of schizophrenia patients using the Illness Perception Questionnaire. *British Journal of Clinical Psychology*, *40*(4), 371–385. doi:10.1348/014466501163869

Barrowclough, C., & Parle, M. (1997). Appraisal, psychological adjustment and expressed emotion in relatives of patients suffering from schizophrenia. *British Journal of Psychiatry*, *170*(JULY), 26–30. doi:10.1192/bjp.171.1.26

Bauer, R., Sterzinger, L., Koepke, F., & Spiessl, H. (2013). Rewards of caregiving and coping strategies of caregivers of patients with mental illness. *Psychiatric Services*, *64*(2), 185–188. doi:10.1176/appi.ps.001212012

Bebbington, P., & Kuipers, L. (1994). The predictive utility of expressed emotion in schizophrenia: an aggregate analysis. *Psychological Medicine*, *24*(3), 707–18. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/7991753

Berglund, N., Vahlne, J. O., & Edman, A. (2003). Family intervention in schizophrenia--impact on family burden and attitude. *Social Psychiatry and Psychiatric Epidemiology*, *38*(3), 116–21. doi:10.1007/s00127-003-0615-6

Bijl, R. V., Cuijpers, P., & Smit, F. (2002). Psychiatric disorders in adult children of parents with a history of psychopathology. *Social Psychiatry and Psychiatric Epidemiology*, *37*(1), 7–12. doi:10.1007/s127-002-8208-8

Bird, V., Premkumar, P., Kendall, T., Whittington, C., Mitchell, J., & Kuipers, E. (2010). Early intervention services, cognitive-behavioural therapy and family intervention in early psychosis: systematic review. *The British Journal of Psychiatry : The Journal of Mental Science*, *197*(5), 350–6. doi:10.1192/bjp.bp.109.074526

Bjelland, I., Dahl, A. A., Haug T. T., & Neckelmann D. (2002) The validity of the Hospital Anxiety and Depression Scale: An updated literature review. *Journal of Psychosomatic Research, 52(2),* 69-77. doi:10.1016/S0022-3999(01)00296-3

Bratiotis, C. (2013). Community hoarding task forces: A comparative case study of five task forces in the United States. *Health and Social Care in the Community*, *21*(3), 245–253. doi:10.1111/hsc.12010

Braun, V., & Clarke, V. (2008). Using thematic analysis in psychology. *Qualitative Research in Psychology*. Retrieved from http://www.tandfonline.com/doi/abs/10.1191/1478088706qp063oa#.VRgStvnF8XU

Broadbent, E., Petrie, K. J., Main, J., & Weinman, J. (2006). The brief illness perception questionnaire. *Journal of Psychosomatic Research*, *60*(6), 631–7. doi:10.1016/j.jpsychores.2005.10.020

Budd, R. J., Oles, G., & Hughes, I. C. T. (1998). The relationship between coping style and burden in the carers of reatives with schizophrenia. *Acta Psychiatrica Scandinavica*, *98*(4), 304–309. doi:10.1111/j.1600-0447.1998.tb10088.x

Büscher, T. P., Dyson, J., & Cowdell, F. (2014). The effects of hoarding disorder on families: An integrative review. *Journal of Psychiatric and Mental Health Nursing*, *21*(6), 491–498. doi:10.1111/jpm.12098

Butzlaff, R. L., & Hooley, J. M. (1998). Expressed Emotion and Psychiatric Relapse. *Archives of General Psychiatry*, *55*(6), 547. doi:10.1001/archpsyc.55.6.547

Carver, C. S. (1997). You want to measure coping but your protocol's too long: consider the brief COPE. *International Journal of Behavioral Medicine*, *4*(1), 92–100. doi:10.1207/s15327558ijbm0401_6

Cechnicki, A., Bielańska, A., Hanuszkiewicz, I., & Daren, A. (2013). The predictive validity of Expressed Emotions (EE) in schizophrenia. A 20-year prospective study. *Journal of Psychiatric Research*, *47*(2), 208–214. doi:10.1016/j.jpsychires.2012.10.004

Chambless, D. L., & Steketee, G. (1999). Expressed emotion and behavior therapy outcome: a prospective study with obsessive-compulsive and agoraphobic outpatients. *Journal of Consulting and Clinical Psychology*, *67*(5), 658–665. doi:10.1037/0022-006X.67.5.658

Chasson, G. S., Carpenter, A., Ewing, J., Gibby, B., & Lee, N. (2014). Empowering families to help a loved one with Hoarding Disorder: Pilot study of Family-As-Motivators training. *Behaviour Research and Therapy*, *63*, 9–16. doi:10.1016/j.brat.2014.08.016

Chen, F., & Greenberg, J. S. (2004). A Positive Aspect of Caregiving: The Influence of Social Support on Caregiving Gains for Family Members of Relatives with Schizophrenia. *Community Mental Health Journal*, *40*(5), 423–435. doi:10.1023/B:COMH.0000040656.89143.82

Cherian, A. V., Pandian, D., Bada Math, S., Kandavel, T., & Janardhan Reddy, Y. C. (2014). Family accommodation of obsessional symptoms and naturalistic outcome of obsessive-compulsive disorder. *Psychiatry Research*, *215*(2), 372–378. doi:10.1016/j.psychres.2013.11.017

Chien, W.-T. T., & Norman, I. (2009). The effectiveness and active ingredients of mutual support groups for family caregivers of people with psychotic disorders: A literature review. *International Journal of Nursing Studies*, *46*(12), 1604–1623. doi:10.1016/j.ijnurstu.2009.04.003

Christenson, J. D., Crane, D. R., Bell, K. M., Beer, A. R., & Hillin, H. H. (2014). Family Interventions and Health Care Costs for Kansas Medicaid Patients with Schizophrenia. *Journal of Marital & Family Therapy*, *40*(3), 272–286. doi:10.1111/jmft.12021

Citron, M., Solomon, P., & Draine, J. (1999). Self-Help Groups for Families of Persons with Mental Illness: Perceived Benefits of Helpfulness. *Community Mental Health Journal*, *35*(1), 15–30. doi:10.1023/A:1018791824546

Cohen, C. A., Colantonio, A., & Vernich, L. (2002). Positive aspects of caregiving: rounding out the caregiver experience. *International Journal of Geriatric Psychiatry*, *17*(2), 184–8. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/11813283

Cooklin, A. (2006). Children of Parents with Mental Illness. *Psychiatry*, *4*(1), 32–35.

Cooklin, A. (2010). "Living upside down": being a young carer of a parent with mental illness. *Advances in Psychiatric Treatment*, *16*(2), 141–146. doi:10.1192/apt.bp.108.006247

Cuijpers, P. (1999). The effects of family interventions on relatives' burden: A meta-analysis. *Journal of Mental Health*, *8*(3), 275–285. doi:10.1080/09638239917436

Cuijpers, P., Hosman, C. M. H., & Munnichs, J. M. A. (1996). Change Mechanisms of Support Groups for Caregivers of Dementia Patients. *International Psychogeriatrics*, *8*(04), 575–587. Retrieved from http://journals.cambridge.org/abstract_S104161029600289X

Damecour, C. L., & Charron, M. (1998). Hoarding: a symptom, not a syndrome. *The Journal of Clinical Psychiatry*, *59*(5), 267–72; quiz 273. Retrieved from http://europepmc.org/abstract/med/9632043

Dimauro, J., Tolin, D. F., Frost, R. O., & Steketee, G. (2013). Do People with Hoarding Disorder Under-Report Their Symptoms? *Journal of Obsessive-Compulsive and Related Disorders*, *2*(2), 130–136. doi:10.1016/j.jocrd.2013.01.002

Drury, H., Ajmi, S., Fernández de la Cruz, L., Nordsletten, A. E., & Mataix-Cols, D. (2014). Caregiver burden, family accommodation, health, and well-being in relatives of individuals with hoarding disorder. *Journal of Affective Disorders*, *159*, 7–14. doi:10.1016/j.jad.2014.01.023

Faul, F., Erdfelder, E., Lang, A.-G., & Buchner, A. (2007). G*Power 3: a flexible statistical power analysis program for the social, behavioral, and biomedical sciences. *Behavior Research Methods*, *39*(2), 175–91. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/17695343

Fortune, D. G., Smith, J. V, & Garvey, K. (2005). Perceptions of psychosis, coping, appraisals, and psychological distress in the relatives of patients with schizophrenia: an exploration using self-regulation theory. *The British Journal of Clinical Psychology / the British Psychological Society*, *44*(Pt 3), 319–31. doi:10.1348/014466505X29198

Frost, R. O., & Gross, R. C. (1993). The hoarding of possessions. *Behaviour Research and Therapy*, *31*(4), 367–381. doi:10.1016/0005-7967(93)90094-B

Frost, R. O., Steketee, G., & Tolin, D. F. (2011). Comorbidity in hoarding disorder. *Depression and Anxiety*, *28*(10), 876–84. doi:10.1002/da.20861

Frost, R. O., Steketee, G., Tolin, D. F., & Renaud, S. (2008). Development and Validation of the Clutter Image Rating. *Journal of Psychopathology and Behavioral Assessment*, *30*(3), 193–203. doi:10.1007/s10862-007-9068-7

Frost, R. O., Steketee, G., & Williams, L. (2000). Hoarding: a community health problem. *Health & Social Care in the Community*, *8*(4), 229–234. doi:10.1046/j.1365-2524.2000.00245.x

Frost, R. O., Steketee, G., Williams, L. F., & Warren, R. (2000). Mood, personality disorder symptoms and disability in obsessive compulsive hoarders: a comparison with clinical and nonclinical controls. *Behaviour Research and Therapy*, *38*(11), 1071–81. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/11060936

Frost, R. O., Steketee, G., Youngren, V. R., & Mallya, G. K. (1999). The Threat of the Housing Inspector: A Case of Hoarding. *Harvard Review of Psychiatry*, *6*(5), 270–278. Retrieved from http://informahealthcare.com/doi/abs/10.3109/10673229909000339?journalCode=hrp

Gaebel, W., Weinmann, S., Sartorius, N., Rutz, W., & McIntyre, J. S. (2005). Schizophrenia practice guidelines: international survey and comparison. *The British Journal of Psychiatry : The Journal of Mental Science*, *187*(3), 248–55. doi:10.1192/bjp.187.3.248

Garety, P. A., Fowler, D. G., Freeman, D., Bebbington, P., Dunn, G., & Kuipers, E. (2008). Cognitive--behavioural therapy and family intervention for relapse prevention and symptom reduction in psychosis: randomised controlled trial. *The British Journal of Psychiatry : The Journal of Mental Science*, *192*(6), 412–23. doi:10.1192/bjp.bp.107.043570

Gilliam, C. M., Norberg, M. M., Villavicencio, A., Morrison, S., Hannan, S. E., & Tolin, D. F. (2011). Group cognitive-behavioral therapy for hoarding disorder : An open trial. *Behaviour Research and Therapy*, *49*(11), 802–807. doi:10.1016/j.brat.2011.08.008

Goddard, E., Macdonald, P., Sepulveda, A. R., Naumann, U., Landau, S., Schmidt, U., & Treasure, J. (2011). Cognitive interpersonal maintenance model of eating disorders: Intervention for carers. *British Journal of Psychiatry*, *199*(3), 225–231. doi:10.1192/bjp.bp.110.088401

Goldberg, D. P., & Hillier, V. F. (1979). A scaled version of the General Health Questionnaire. *Psychological Medicine*, *9*, 139–145. doi:10.1017/S0033291700021644

Goldberg, D. P., & Williams, P. (1988). *A user's guide to the General Health Questionnaire.* Windsor, UK: NFER-Nelson.

Grisham, J. R., Frost, R. O., Steketee, G., Kim, H.-J., & Hood, S. (2006). Age of onset of compulsive hoarding. *Journal of Anxiety Disorders*, *20*(5), 675–86. doi:10.1016/j.janxdis.2005.07.004

Grisham, J. R., Steketee, G., & Frost, R. O. (2008). Interpersonal problems and emotional intelligence in compulsive hoarding. *Depression and Anxiety*, *25*(9), E63–E71. doi:10.1002/da.20327

Gutiérrez-Maldonado, J., Caqueo-Urízar, A., & Ferrer-García, M. (2009). Effects of a psychoeducational intervention program on the attitudes and health perceptions of relatives of patients with schizophrenia. *Social Psychiatry and Psychiatric Epidemiology*, *44*(5), 343–8. doi:10.1007/s00127-008-0451-9

Gutiérrez-Maldonado, J., Caqueo-Urízar, A., & Kavanagh, D. J. (2005). Burden of care and general health in families of patients with schizophrenia. *Social Psychiatry and Psychiatric Epidemiology*, *40*(11), 899–904. doi:10.1007/s00127-005-0963-5

Hooley, J. M., & Parker, H. A. (2006). Measuring expressed emotion: an evaluation of the shortcuts. *Journal of Family Psychology*, *20*(3), 386–96. doi:10.1037/0893-3200.20.3.386

IBM Corp. (2013). IBM SPSS Statistics for Windows, Version 22.0. Armonk, NY: IBM Corp.

Iervolino, A. C., Ph, D., Perroud, N., Fullana, M. A., Guipponi, M., Cherkas, L., … Mataix-cols, D. (2009). Prevalence and Heritability of Compulsive Hoarding : A Twin Study. *American Journal of Psychiatry*, *166*(October), 1156–1161.

Jansen, J. E., Gleeson, J., & Cotton, S. (2015). Towards a better understanding of caregiver distress in early psychosis: A systematic review of the psychological factors involved. *Clinical Psychology Review*, *35*, 56–66. doi:10.1016/j.cpr.2014.12.002

Jones, D. W. (2004). Families and serious mental illness: Working with loss and ambivalence. *British Journal of Social Work*, *34*(7), 961–979. doi:10.1093/bjsw/bch123

Joyce, J., Leese, M., & Szmukler, G. (2000). The Experience of Caregiving Inventory: further evidence. *Social Psychiatry and Psychiatric Epidemiology*, *35*(4), 185–189. doi:10.1007/s001270050202

Kalra, H., Kamath, P., Trivedi, J. K., & Janca, A. (2008). Caregiver burden in anxiety disorders. *Current Opinion in Psychiatry*, *21*(1), 70–73. doi:10.1097/YCO.0b013e3282f2dfca

Kate, N., Grover, S., Kulhara, P., & Nehra, R. (2013). Positive aspects of caregiving and its correlates in caregivers of schizophrenia: A study from North India. *East Asian Archives of Psychiatry*, *23*(2), 45–55.

Khan, A., Thompson, C., Onwumere, J. (2015) *The impact of caregiving in Hoarding Disorder: An in-depth evaluation of a psycho-educational group intervention for relatives of individuals with Hoarding Disorder.* King's College London.

Kim, H.-J., Steketee, G., & Frost, R. O. (2001). Hoarding by Elderly People. *Health & Social Work*, *26*(3), 176–184. doi:10.1093/hsw/26.3.176

Kuipers, E., Bebbington, P., Dunn, G., Fowler, D., Freeman, D., Watson, P., … Garety, P. (2006). Influence of carer expressed emotion and affect on relapse in non-affective psychosis. *British Journal of Psychiatry*, *188*(FEB.), 173–179. doi:10.1192/bjp.bp.104.007294

Kuipers, E., Onwumere, J., & Bebbington, P. (2010). Cognitive model of caregiving in psychosis. *The British Journal of Psychiatry : The Journal of Mental Science*, *196*(4), 259–65. doi:10.1192/bjp.bp.109.070466

Kuipers, E., & Raune, D. (2000). The early development of expressed emotion and burden in the families of first-onset psychosis. In M. Birchwood, D. Fowler, & C. Jackson (Eds.), *Early Intervention in psychosis* (pp. 128–140). Chichester, UK: John Wiley & Sons Ltd.

Kulhara, P. (2012). Positive aspects of caregiving in schizophrenia: A review. *World Journal of Psychiatry*, *2*(3), 43. doi:10.5498/wjp.v2.i3.43

Kyriacou, O., Treasure, J., & Schmidt, U. (2008). Expressed emotion in eating disorders assessed via self-report: an examination of factors associated with expressed emotion in carers of people with anorexia nervosa in comparison to control families. *The International Journal of Eating Disorders*, *41*(1), 37–46. doi:10.1002/eat.20469

Lebowitz, E. R., Panza, K. E., Su, J., & Bloch, M. H. (2012). Family accommodation in obsessive-compulsive disorder. *Expert Review of Neurotherapeutics*, *12*(2), 229–38. doi:10.1586/ern.11.200

Leff, J. P., & Vaughn, C. E. (1985). *Expressed emotion in families*. New York: Guildford Press.

Lobban, F., Barrowclough, C., & Jones, S. (2005). Assessing cognitive representations of mental health problems. I. The illness perception questionnaire for schizophrenia. *The British Journal of Clinical Psychology / the British Psychological Society*, *44*(Pt 2), 147–62. doi:10.1348/014466504X19497

Lobban, F., Postlethwaite, A., Glentworth, D., Pinfold, V., Wainwright, L., Dunn, G., … Haddock, G. (2013). A systematic review of randomised controlled trials of interventions reporting outcomes for relatives of people with psychosis. *Clinical Psychology Review*, *33*(3), 372–382. doi:10.1016/j.cpr.2012.12.004

Lowenstein, J. A., Butler, D. W., & Ashcroft, K. (2010). The efficacy of a cognitively orientated carers group in an early intervention in psychosis service--a pilot study. *Journal of Psychiatric and Mental Health Nursing*, *17*(7), 628–35. doi:10.1111/j.1365-2850.2010.01564.x

Lucksted, A., McFarlane, W., Downing, D., & Dixon, L. (2012). Recent developments in family psychoeducation as an evidence-based practice. *Journal of Marital and Family Therapy*, *38*(1), 101–21. doi:10.1111/j.1752-0606.2011.00256.x

Magaña, A. B., Goldstein, M. J., Karno, M., Miklowitz, D. J., Jenkins, J., & Falloon, I. R. H. (1986). A brief method for assessing expressed emotion in relatives of psychiatric patients. *Psychiatry Research*, *17*(3), 203–212. doi:10.1016/0165-1781(86)90049-1

Magaña, S. M., Ramírez García, J. I., Hernández, M. G., & Cortez, R. (2007). Psychological Distress Among Latino Family Caregivers of Adults With Schizophrenia: The Roles of Burden and Stigma. *Psychiatric Services*, *58*(3), 378–384. Retrieved from http://ps.psychiatryonline.org/doi/full/10.1176/ps.2007.58.3.378

Magliano, L., Tosini, P., Guarneri, M., Marasco, C., & Catapano, F. (1996). Burden on the families of patients with obsessive-compulsive disorder: a pilot study. *European Psychiatry : The Journal of the Association of European Psychiatrists*, *11*(4), 192–7. doi:10.1016/0924-9338(96)88390-8

Maheswaran, H., Weich, S., Powell, J., & Stewart-Brown, S. (2012). Evaluating the responsiveness of the Warwick Edinburgh Mental Well-Being Scale (WEMWBS): group and individual level analysis. *Health and Quality of Life Outcomes*, *10*(1), 156. doi:10.1186/1477-7525-10-156

Mak, W. W. S., & Cheung, R. Y. M. (2012). Psychological distress and subjective burden of caregivers of people with mental illness: the role of affiliate stigma and face concern. *Community Mental Health Journal*, *48*(3), 270–4. doi:10.1007/s10597-011-9422-9

Malla, A. K., Kazarian, S. S., Barnes, S., & Cole, J. D. (1991). Validation of the Five Minute Speech Sample in measuring expressed emotion. *Canadian Journal of Psychitatry*, *36*, 297–99.

Manassis, K., Bradley, S., Goldberg, S., Hood, J., & Swinson, R. P. (1994). Attachment in mothers with anxiety disorders and their children. *Journal of the American Academy of Child and Adolescent Psychiatry*, *33*(8), 1106–13. - doi:10.1097/00004583-199410000-00006

Mangalore, R., & Knapp, M. (2007). Cost of schizophrenia in England. *The Journal of Mental Health Policy and Economics*, *10*(1), 23–41. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/17417045

Marlatt, G. A. (1998). Highlights of harm reduction: A personal report from the first national harm reduction conference in the United States. In G. A. Marlatt (Ed.), *Harm reduction: Pragmatic strategies for managing high risk behaviors.* New York: Guildford.

Marley, A., Thompson, C., & Onwumere, J. (2015). *The wider impact of hoarding disorder: a qualitative study exploring the experiences of carers of individuals with hoarding disorder, and developing an information leaflet*. King's College London.

Mataix-Cols, D., Billotti, D., Fernández de la Cruz, L., & Nordsletten, A. E. (2013). The London field trial for hoarding disorder. *Psychological Medicine*, *43*(4), 837–47. doi:10.1017/S0033291712001560

McFarlane, W. R., Dixon, L., Lukens, E., & Lucksted, A. (2003). Family psychoeducation and schizophrenia: a review of the literature. *Journal of Marital and Family Therapy*, *29*(2), 223–245. doi:10.1111/j.1752-0606.2003.tb01202.x

McGuire, J. F., Kaercher, L., Park, J. M., & Storch, E. A. (2013). Hoarding in the Community: A Code Enforcement and Social Service Perspective. *Journal of Social Service Research*, *39*(3), 335–344. doi:10.1080/01488376.2013.770813

Medard, E., & Kellett, S. (2014). The Role of Adult Attachment and Social Support in Hoarding Disorder. *Behavioural and Cognitive Psychotherapy*, *42*, 1–5. doi:10.1017/S1352465813000659

Miklowitz, D. J. (2006). A review of evidence-based psychosocial interventions for bipolar disorder. *Journal of Clinical Psychiatry*, *67*(SUPPL. 11), 28–33.

Miklowitz, D. J., O'Brien, M. P., Schlosser, D. A., Addington, J., Candan, K. A., Marshall, C., … Cannon, T. D. (2014). Family-focused treatment for adolescents and young adults at high risk for psychosis: results of a randomized trial. *Journal of the American Academy of Child and Adolescent Psychiatry*, *53*(8), 848–58. doi:10.1016/j.jaac.2014.04.020

Miller, F., Dworkin, J., Ward, M., & Barone, D. (1990). A Preliminary Study of Unresolved Grief in Families of Seriously Mentally Ill Patients. *Psychiatric Services*, *41*(12), 1321–1325. doi:10.1176/ps.41.12.1321

Mowbray, C. T., Bybee, D., Oyserman, D., MacFarlane, P., & Bowersox, N. (2006). Psychosocial outcomes for adult children of parents with severe mental illnesses: demographic and clinical history predictors. *Health & Social Work*, *31*(2), 99–108. doi:10.1093/hsw/31.2.99

Mueller, A., Mitchell, J. E., Crosby, R. D., Glaesmer, H., & de Zwaan, M. (2009). The prevalence of compulsive hoarding and its association with compulsive buying in a German population-based sample. *Behaviour Research and Therapy*, *47*(8), 705–709. doi:10.1016/j.brat.2009.04.005

NICE. (2009). Schizophrenia: Core Interventions in the Treatment and Management of Schizophrenia in Adults in Primary and Secondary Care. Retrieved from Available at www.nice.org.uk/CG82

NICE. (2014). *Psychosis and schizophrenia in adults: treatment and management. NICE clinical guideline 179*. Retrieved from https://www.nice.org.uk/guidance/cg178/chapter/1-Recommendations

Nordsletten, A. E., Fernández de la Cruz, L., Drury, H., Ajmi, S., Saleem, S., & Mataix-Cols, D. (2014). The Family Impact Scale for Hoarding (FISH): Measure development and initial validation. *Journal of Obsessive-Compulsive and Related Disorders*, *3*(1), 29–34. doi:10.1016/j.jocrd.2013.10.003

Nordsletten, A. E., Fernández de la Cruz, L., Pertusa, A., Reichenberg, A., Hatch, S. L., Mataix-Cols, D., … Mataix-Cols, D. (2013). The Structured Interview for Hoarding Disorder (SIHD): Development, usage and further validation. *Journal of Obsessive-Compulsive and Related Disorders*, *2*(3), 346–350. doi:10.1016/j.jocrd.2013.06.003

Nordsletten, A. E., Reichenberg, A., Hatch, S. L., de la Cruz, L. F., Pertusa, A., Hotopf, M., & Mataix-Cols, D. (2013). Epidemiology of hoarding disorder. *The British Journal of Psychiatry : The Journal of Mental Science*, *203*(6), 445–52. doi:10.1192/bjp.bp.113.130195

Office for National Statistics. (2012). *2011 Census: Ethnic group, local authorities in the United Kingdom*. *2011 Census*. Retrieved April 20, 2015, from http://www.ons.gov.uk/ons/rel/census/2011-census/key-statistics-for-local-authorities-in-england-and-wales/rpt-ethnicity.html

Oldridge, M. L., & Hughes, I. C. (1992). Psychological well-being in families with a member suffering from schizophrenia. An investigation into long-standing problems. *The British Journal of Psychiatry*, *161*(2), 249–251. doi:10.1192/bjp.161.2.249

Onwumere, J., Kuipers, E., Bebbington, P., Dunn, G., Fowler, D., Freeman, D., … Garety, P. (2008). Caregiving and illness beliefs in the course of psychotic illness. *Canadian Journal of Psychiatry. Revue Canadienne de Psychiatrie*, *53*(7), 460–8. Retrieved from http://europepmc.org/abstract/med/18674404

Onwumere, J., Kuipers, E., Bebbington, P., Dunn, G., Freeman, D., Fowler, D., & Garety, P. (2011). Coping styles in carers of people with recent and long-term psychosis. *The Journal of Nervous and Mental Disease*, *199*(6), 423–4. doi:10.1097/NMD.0b013e31821ccb07

Park, J. M., Lewin, A. B., & Storch, E. a. (2014). Adult offspring perspectives on parental hoarding behaviors. *Psychiatry Research*, *220*, 328–334. doi:10.1016/j.psychres.2014.07.061

Patterson, P., Birchwood, M., & Cochrane, R. (2005). Expressed emotion as an adaptation to loss: prospective study in first-episode psychosis. *The British Journal of Psychiatry. Supplement*, *48*(48), s59–64. doi:10.1192/bjp.187.48.s59

Perlick, D. A., Hohenstein, J. M., Clarkin, J. F., Kaczynski, R., & Rosenheck, R. A. (2005). Use of mental health and primary care services by caregivers of patients with bipolar disorder: a preliminary study. *Bipolar Disorders*, *7*(2), 126–35. doi:10.1111/j.1399-5618.2004.00172.x

Perlick, D. A., Rosenheck, R. A., Miklowitz, D. J., Kaczynski, R., Link, B., Ketter, T., … Sachs, G. (2008). Caregiver burden and health in bipolar disorder: a cluster analytic approach. *The Journal of Nervous and Mental Disease*, *196*(6), 484–91. doi:10.1097/NMD.0b013e3181773927

Pertusa, A., Fullana, M. A., Singh, S., Alonso, P., Menchón, J. M., & Mataix-Cols, D. (2008). Compulsive hoarding: OCD symptom, distinct clinical syndrome, or both? *The American Journal of Psychiatry*, *165*(10), 1289–98. doi:10.1176/appi.ajp.2008.07111730

Petrakis, M., Oxley, J., & Bloom, H. (2012). Carer psychoeducation in first-episode psychosis: Evaluation outcomes from a structured group programme. *International Journal of Social Psychiatry*, *59*(4), 391–7. doi:10.1177/0020764012438476

Pharoah, F., Mari, J., Rathbone, J., & Wong, W. (2010). Family intervention for schizophrenia. *The Cochrane Database of Systematic Reviews*, (12), CD000088. doi:10.1002/14651858.CD000088.pub2

Pilowsky, D. J., Wickramaratne, P., Nomura, Y., & Weissman, M. M. (2006). Family discord, parental depression, and psychopathology in offspring: 20-year follow-up. *Journal of the American Academy of Child and Adolescent Psychiatry*, *45*(4), 452–460. doi:10.1097/01.chi.0000198592.23078.8d

Pinsof, W. M., & Wynne, L. C. (1995). The efficacy of marital and family therapy: An empirical overview, conclusions, and recommendations. *Journal of Marital and Family Therapy*, *21*(4), 585–613. doi:10.1111/j.1752-0606.1995.tb00179.x

Ramos-Cerqueira, A. T. D. A., Torres, A. R., Torresan, R. C., Negreiros, A. P. M., & Vitorino, C. N. (2008). Emotional burden in caregivers of patients with obsessive-compulsive disorder. *Depression and Anxiety*, *25*(12), 1020–1027. doi:10.1002/da.20431

Raune, D., Kuipers, E., & Bebbington, P. E. (2004). Expressed emotion at first-episode psychosis : investigating a carer appraisal model. *British Journal of Psychiatry*, *184*, 321–326. doi:10.1192/bjp.184.4.321

Renshaw, K. D., Steketee, G., & Chambless, D. L. (2005). Involving family members in the treatment of OCD. *Cognitive Behaviour Therapy*, *34*(3), 164–75. doi:10.1080/16506070510043732

Sampson, J. M. (2013). The Lived Experience of Family Members of Persons Who Compulsively Hoard: A Qualitative Study. *Journal of Marital and Family Therapy*, *39*(3), 388–402. doi:10.1111/j.1752-0606.2012.00315.x

Sampson, J. M., Yeats, J. R., & Harris, S. M. (2012). An Evaluation of an Ambiguous Loss Based Psychoeducational Support Group for Family Members of Persons Who Hoard: A Pilot Study. *Contemporary Family Therapy*, *34*(4), 566–581. doi:10.1007/s10591-012-9214-6

Samuels, J. F., Bienvenu, O. J., Grados, M. A., Cullen, B., Riddle, M. A., Liang, K.-Y., … Nestadt, G. (2008). Prevalence and correlates of hoarding behavior in a

community-based sample. *Behaviour Research and Therapy*, *46*(7), 836–44. doi:10.1016/j.brat.2008.04.004

Saxena, S., Ayers, C. R., Maidment, K. M., Vapnik, T., Wetherell, J. L., & Bystritsky, A. (2011). Quality of life and functional impairment in compulsive hoarding. *Journal of Psychiatric Research*, *45*(4), 475–80. doi:10.1016/j.jpsychires.2010.08.007

Scazufca, M., & Kuipers, E. (1996). Links between expressed emotion and burden of care in relatives of patients with schizophrenia. *The British Journal of Psychiatry*, *168*(5), 580–587. doi:10.1192/bjp.168.5.580

Sepulveda, A. R., Kyriacou, O., & Treasure, J. (2009). Development and validation of the accommodation and enabling scale for eating disorders (AESED) for caregivers in eating disorders. *BMC Health Services Research*, *9*, 171. doi:10.1186/1472-6963-9-171

Sin, J., & Norman, I. (2013). Psychoeducational interventions for family members of people with schizophrenia: A mixed-method systematic review. *Journal of Clinical Psychiatry*, *74*(12). doi:10.4088/JCP.12r08308

Smith, L., Onwumere, J., Craig, T., McManus, S., Bebbington, P., & Kuipers, E. (2014). Mental and physical illness in caregivers: results from an English national survey sample. *The British Journal of Psychiatry : The Journal of Mental Science*. doi:10.1192/bjp.bp.112.125369

Stansfeld, S., Smuk, M., Onwumere, J., Clark, C., Pike, C., Mcmanus, S., … Bebbington, P. (2014). Stressors and common mental disorder in informal carers e An analysis of the English Adult Psychiatric Morbidity Survey 2007. *Social Science & Medicine (1982)*, *120*, 190–8. doi:10.1016/j.socscimed.2014.09.025

Stewart-Brown, S., & Janmohamed, K. (2008). *Warwick-Edinburgh Mental Well-being Scale (WEMWBS) User Guide*. Warwick. doi:http://www.healthscotland.com/documents/2702.aspx

Storch, E. A., Geffken, G. R., Merlo, L. J., Jacob, M. L., Murphy, T. K., Goodman, W. K., … Grabill, K. (2007). Family accommodation in pediatric obsessive-compulsive disorder. *Journal of Clinical Child and Adolescent Psychology*, *36*(2), 207–216. doi:10.1080/15374410701277929

Storch, E. A., Geffken, G. R., Merlo, L. J., Mann, G., Duke, D., Munson, M., … Goodman, W. K. (2007). Family-based cognitive-behavioral therapy for pediatric obsessive-compulsive disorder: comparison of intensive and weekly approaches. *Journal of the American Academy of Child and Adolescent Psychiatry*. doi:10.1097/chi.0b013e31803062e7

Storch, E. A., Muroff, J., Lewin, A. B., Geller, D., Ross, A., McCarthy, K., … Steketee, G. (2011). Development and preliminary psychometric evaluation of the Children's

Saving Inventory. *Child Psychiatry and Human Development*, *42*(2), 166–82. doi:10.1007/s10578-010-0207-0

Szmukler, G. I., Burgess, P., Herrman, H., Benson, a, Colusa, S., & Bloch, S. (1996). Caring for relatives with serious mental illness: the development of the Experience of Caregiving Inventory. *Social Psychiatry and Psychiatric Epidemiology*, *31*(3-4), 137–48. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/8766459

Szmukler, G., Kuipers, E., Joyce, J., Harris, T., Leese, M., Maphosa, W., & Staples, E. (2003). An exploratory randomised controlled trial of a support programme for carers of patients with a psychosis. *Social Psychiatry and Psychiatric Epidemiology*, *38*(8), 411–8. doi:10.1007/s00127-003-0652-1

Tennant, R., Hiller, L., Fishwick, R., Platt, S., Joseph, S., Weich, S., … Stewart-Brown, S. (2007). The Warwick-Edinburgh Mental Well-being Scale (WEMWBS): development and UK validation. *Health and Quality of Life Outcomes*, *5*(1), 63. doi:10.1186/1477-7525-5-63

Timpano, K. R., Exner, C., Glaesmer, H., Reif, W., Keshavia, A., Brahler, E., & Wilhelm, S. (2011). The Epidemiology of the Proposed DSM-5 Hoarding Disorder: Exploration of the Acquisition Specifier, Associated Features, and Distress. *The Journal of Clinical Psychiatry*, *72*(6), 780–786. Retrieved from http://cat.inist.fr/?aModele=afficheN&cpsidt=24310573

Tolin, D. F., Fitch, K. E., Frost, R. O., & Steketee, G. (2010). Family Informants' Perceptions of Insight in Compulsive Hoarding. *Cognitive Therapy and Research*, *34*(1), 69–81. doi:10.1007/s10608-008-9217-7

Tolin, D. F., Frost, R. O., & Steketee, G. (2007). An Open Trial of Cognitive-Behavioural Therapy for Compulsive Hoarding. *Behaviour Research and Therapy*, *45*(7), 1461–1470.

Tolin, D. F., Frost, R. O., & Steketee, G. (2010). A brief interview for assessing compulsive hoarding: the Hoarding Rating Scale-Interview. *Psychiatry Research*, *178*(1), 147–52. doi:10.1016/j.psychres.2009.05.001

Tolin, D. F., Frost, R. O., Steketee, G., & Fitch, K. E. (2008). Family burden of compulsive hoarding: results of an internet survey. *Behaviour Research and Therapy*, *46*(3), 334–44. doi:10.1016/j.brat.2007.12.008

Tolin, D. F., Frost, R. O., Steketee, G., Gray, K. D., & Fitch, K. E. (2008). The economic and social burden of compulsive hoarding. *Psychiatry Research*, *160*(2), 200–11. doi:10.1016/j.psychres.2007.08.008

Tolin, D. F., Frost, R. O., Steketee, G., & Muroff, J. (2015). Cognitive Behavioral Therapy for Hoarding Disorder: a Meta-Analysis. *Depression and Anxiety*, *32*, 158–166. doi:10.1002/da.22327

Tompkins, M. A. (2011). Working with families of people who hoard: a harm reduction approach. *Journal of Clinical Psychology*, *67*(5), 497–506. doi:10.1002/jclp.20797

Treasure, J., Murphy, T. K., Szmukler, G., Todd, G., Gavan, K., & Joyce, J. (2001). The experience of caregiving for severe mental illness : a comparison between anorexia nervosa and psychosis. *Social Psychiatry and Psychiatric Epidemiology*, *36*, 343–347.

Treasure, J., Sepulveda, A. R., MacDonal, P., Whitaker, W., Lopez, C., Zabala, M., … Todd, G. (2008). The Assessment of the Family of People with Eating Disorders. *European Eating Disorders Review*, *16*, 247–255. doi:10.1002/erv

Turner, K., Steketee, G., & Nauth, L. (2010). Treating Elders With Compulsive Hoarding: A Pilot Program. *Cognitive and Behavioral Practice*, *17*(4), 449–457. doi:10.1016/j.cbpra.2010.04.001

Van IJzendoorn, M. (1995). Adult attachment representations, parental responsiveness, and infant attachment: A meta-analysis on the predictive validity of the Adult Attachment Interview. *Psychological Bulletin*, *117*(3), 387–403.

Veltman, A., Cameron, J., & Stewart, D. E. (2002). The experience of providing care to relatives with chronic mental illness. *The Journal of Nervous and Mental Disease*, *190*(2), 108–114. doi:10.1097/00005053-200202000-00008

Wahl, O. F., & Harman, C. R. (1989). Family Views of Stigma. *Schizophrenia Bulletin*, *15*(1), 131–139. doi:10.1093/schbul/15.1.131

Whiteford, H. A., Degenhardt, L., Rehm, J., Baxter, A. J., Ferrari, A. J., Erskine, H. E., … Vos, T. (2013). Global burden of disease attributable to mental and substance use disorders: findings from the Global Burden of Disease Study 2010. *Lancet*, *382*(9904), 1575–86. doi:10.1016/S0140-6736(13)61611-6

Wilbram, M., Kellett, S., & Beail, N. (2008). Compulsive hoarding: a qualitative investigation of partner and carer perspectives. *The British Journal of Clinical Psychology / the British Psychological Society*, *47*(Pt 1), 59–73. doi:10.1348/014466507X240740

Yesufu-Udechuku, a., Harrison, B., Mayo-Wilson, E., Young, N., Woodhams, P., Shiers, D., … Kendall, T. (2015). Interventions to improve the experience of caring for people with severe mental illness: systematic review and meta-analysis. *The British Journal of Psychiatry*, *206*(4), 268–274. doi:10.1192/bjp.bp.114.147561

Zigmond, A. S., & Snaith, R. P. (1983). The Hospital Anxiety and Depression Scale. *Acta Psychiatrica Scandinavica*, *67*(6), 361–70. doi:10.1111/j.1600-0447.1983.tb09716.x

6. Appendices

Appendix 1: Diagnostic criteria for Hoarding Disorder (from DSM-5; APA, 2013)

A. Persistent difficulty discarding or parting with possessions, regardless of their actual value.

B. This difficulty is due to a perceived need to save the items and distress associated with discarding them

C. The symptoms result in the accumulation of possessions that congest and clutter active living areas and substantially compromise their intended use. If living areas are uncluttered, it is only because of the interventions of third parties (e.g. family members, cleaners, authorities)

D. The hoarding causes clinically significant distress or impairment in social, occupational or other important areas of functioning (including maintaining a safe environment for self and others).

E. The hoarding is not attributable to another medical condition (e.g. brain injury, cerebrovascular disease, Prader–Willi syndrome).

F. The hoarding is not better accounted for by the symptoms of another DSM-5 disorder (e.g. hoarding due to obsessions in obsessive–compulsive disorder, decreased energy in major depressive disorder, delusions in schizophrenia or another psychotic disorder, cognitive deficits in dementia, restricted interests in autism spectrum disorder).

Appendix 2: Ethical approval letter from King's College London Psychiatry, Nursing and Midwifery Research Ethics Committee.

Claire Thompson
3rd Floor, Addiction Sciences Building
Institute of Psychiatry
King's College London
4 Windsor Walk
London
SE5 8AF

17th January 2014

Dear Claire Thompson

PNM/13/14-28 The impact of caregiving in Hoarding Disorder: Piloting a brief psychoeducational group for relatives of hoarders

Review Outcome: Full Approval

Thank you for sending in the amendments/clarifications requested to the above project. I am pleased to inform you that these meet the requirements of the PNM RESC and therefore that full approval is now granted with the following provisos:

1. Section 7.2 and Information Sheets: Please specify an exact date for withdrawal of participant's data.

2. Information Sheet: Please replace the title of the paragraph 'What will happen to me if I take part' to something like 'What taking part with involve for me'

Note that you do not need to submit a response to the above proviso, however it is a condition of the approval granted by the PNM RESC that the proviso is carried out prior to the study commencing. If the proviso is not adhered to, the approval granted by the PNM RESC would no longer be valid. Should you have any queries on this please do not hesitate to contact the Research Ethics Office.

Please ensure that you follow all relevant guidance as laid out in the King's College London Guidelines on Good Practice in Academic Research (http://www.kcl.ac.uk/college/policyzone/index.php?id=247)

For your information ethical approval is granted until **14th January 2017**. If you need approval beyond this point you will need to apply for an extension to approval at least two weeks prior to this explaining why the extension is needed, (please note however that a full re-application will not be necessary unless the protocol has changed). You should also note that if your approval is for one year, you will not be sent a reminder when it is due to lapse.

Ethical approval is required to cover the duration of the research study, up to the conclusion of

the research. The conclusion of the research is defined as the final date or event detailed in the study description section of your approved application form (usually the end of data collection when all work with human participants will have been completed), not the completion of data analysis or publication of the results. For projects that only involve the further analysis of pre-existing data, approval must cover any period during which the researcher will be accessing or evaluating individual sensitive and/or un-anonymised records. Note that after the point at which ethical approval for your study is no longer required due to the study being complete (as per the above definitions), you will still need to ensure all research data/records management and storage procedures agreed to as part of your application are adhered to and carried out accordingly.

If you do not start the project within three months of this letter please contact the Research Ethics Office.

Should you wish to make a modification to the project or request an extension to approval you will need approval for this and should follow the guidance relating to modifying approved applications:
http://www.kcl.ac.uk/innovation/research/support/ethics/applications/modifications.aspx
The circumstances where modification requests are required include the addition/removal of participant groups, additions/removal/changes to research methods, asking for additional data from participants, extensions to the ethical approval period. Any proposed modifications should only be carried out once full approval for the modification request has been granted.

Any unforeseen ethical problems arising during the course of the project should be reported to the approving committee/panel. In the event of an untoward event or an adverse reaction a full report must be made to the Chair of the approving committee/review panel within one week of the incident.

Please would you also note that we may, for the purposes of audit, contact you from time to time to ascertain the status of your research.

If you have any query about any aspect of this ethical approval, please contact your panel/committee administrator in the first instance (http://www.kcl.ac.uk/innovation/research/support/ethics/contact.aspx). We wish you every success with this work.

With best wishes

Yours sincerely,

Annah Whyton

Research Support Assistant

INFORMATION SHEET FOR PARTICIPANTS
(PART 1)

REC Reference Number **PNM/13/14-28**

KING'S College LONDON

The Experience of Caring in Hoarding (ECHO) – Study A

Version: 1

We would like to invite you to participate in this research project. You should only participate if you want to; choosing not to take part will not disadvantage you in any way. Before you decide whether you want to take part, it is important for you to understand why the research is being done and what your participation will involve. Please take time to read the following information carefully and discuss it with others if you wish. Please ask the researcher if there is anything that is not clear or if you would like more information.

What is the purpose of the study?

The aim of this study is to improve our understanding of the experiences faced by relatives (carers) of an individual who hoards. It will focus on how people cope with having a relative who hoards and how the relative's hoarding affects them. The results will help us to understand the impact hoarding has on relatives and help us to improve services we offer people who hoard and their families.

Why have I been chosen?

We are inviting all relatives (carers) of people who hoard to take part in the research and we hope at least 40 people will take part. We are working with different organisations and services who might have contact with the carers of adults who hoard. For this project, a carer may be a child, spouse, partner, sibling or friend who is in close and regular contact with the person who hoards.

Do I have to take part?

No. It is entirely up to you to decide whether or not to take part. If you decide to take part, you are free to withdraw from the research *at any time* and *without giving a reason*. Your decision about this will not affect the current or future involvement you may have with the research team or health

professionals or any care that your relative might currently receive or access in the future.

When and where will the study take place?

You can fill in questionnaires online or we can post these out to you. There will also be a short telephone interview with the researcher. If you prefer the researcher can arrange to meet with you face-to-face, if you live in the London area.

What will taking part involve for me?

- If you are happy to take part and the research team has answered any questions you have, you will be given this information sheet to keep and asked to sign a consent form.

- If you agree to take part, you will be asked to fill in some questionnaires which look at the experience of having a relative who hoards. These will include asking you about: the level of severity of your relatives hoarding, your views on their hoarding problem and the impact it has on your well being and health and your relationship with the person who hoards. The questionnaires and interview should take up to 1 ½ hours at the most.

- If you agree to take part you will be asked whether you are happy to be contacted about participation in future studies. Your participation in this study will not be affected should you choose not to be re-contacted.

- If you live in the London area we may also invite you to participate in another part of the study we are running currently in which we are running a support group for relatives of people who hoard. There is no obligation to take part in this second part of the study and a separate information sheet and consent form will be provided for this.

- We would also like to audio-record the interview. These recordings will be stored securely and deleted once we have entered the data.

- You may withdraw from the research project at any time and can withdraw your information from the project up to one month after taking part.

- To thank you for your time in completing the questionnaires you will receive £10 in cash or by cheque. If you live outside of the UK we can reimburse you via a £10 Amazon voucher.

What are the possible benefits of taking part?

We do not expect participating in the study to have any direct benefits for you, although some people report finding the chance to talk about their experiences useful and interesting. Your participation will contribute to our knowledge in this area and help to develop interventions and support for people with similar experiences in the future. We can provide you with a copy of the final report.

What are the possible disadvantages of taking part?

Talking about your worries and experiences can be upsetting for some people. If you have been affected by issues raised by your participation in the study please speak to the research team on the contact details below.

Contact details

If you have any questions or require more information about this study, please contact the researcher using the following contact details: Claire Thompson, Clinical Psychologist in Training, ASB, Institute of Psychiatry, 4 Windsor Walk, London, SE5 8AZ. Email: Claire.c.thompson@kcl.ac.uk, Tel: 0207 848 0733

If the study has harmed you in any way, please contact: Dr Juliana Onwumere, Department of Psychology, Henry Wellcome Building, Institute of Psychiatry, 16 De Crespigny Park, London, SE5 8AF Juliana.1.Onwumere@kcl.ac.uk, Tel: 020 7848 0197

Part 2

Will my taking part in the study be kept confidential?

All the information recorded will be strictly confidential and kept in accordance with the Data Protection Act 1998, and used only by clinicians and researchers working within the research team. The data will be anonymised and will be identifiable only by a number and not by your name. We would only ever break this confidentiality in the case that you revealed something which caused us to be concerned for your safety or the safety of others, in which case we would inform you first and may need to contact appropriate services.

The audio recordings and questionnaires you complete will be kept in a locked cabinet at the research institution and only named researchers will have access to them. Your name will be kept separately with the number on a database and on paper so that we can identify your data in the future if we need to, for example, if you decide you no longer want to be part of the study. Your details will be kept for up to 4 years, and then will be confidentially destroyed. We will keep a completely anonymised copy of the database indefinitely, from which you will not be able to be identified at all.

What will happen to the results of the study?

When the study is completed, we intend to publish the results in order to help other people working with family members of people who hoard, but your information would be completely confidential and you would not be named in the paper. We will also write to all participants and inform you of the results and offer you a copy of the report if you would like it.

Who has reviewed this study?

This study has been reviewed by Psychiatry, Nursing and Midwifery (PNM) Research Ethics Subcommittee (RESC) at King's College London, REC reference number: PNM/13/14-28

You will receive a copy of this information sheet and the signed consent section if you decide to participate.

Thank you for considering taking part in our study.

CONSENT FORM FOR PARTICIPANTS IN RESEARCH STUDIES

Please complete this form after you have read the Information Sheet and/or listened to an explanation about the research.

Title of Study: _ECHO Study_____

King's College Research Ethics Committee Ref:___ PNM/13/14-28 _____

Thank you for considering taking part in this research. The person organising the research must explain the project to you before you agree to take part. If you have any questions arising from the Information Sheet or explanation already given to you, please ask the researcher before you decide whether to join in. You will be given a copy of this Consent Form to keep and refer to at any time.

- I confirm that I have read the information sheet dated for the above study. I have had the opportunity to consider the information and ask questions. ☐
- I understand my participation is voluntary, and that if I decide at any time during the research that I no longer wish to participate in this project, I can notify the researchers involved and withdraw from it immediately without giving any reason. ☐
- I understand that I may withdraw my data from the study up to one month after I take part. Withdraw data up to this date _____ ☐
- I consent to the processing of my personal information for the purposes explained to me. I understand that such information will be handled in accordance with the terms of the UK Data Protection Act 1998. ☐
- I understand that confidentiality and anonymity will be maintained and it will not be possible to identify me in any publications. ☐
- I consent to my interview being audio recorded and understand this will be deleted no later than 1 year after the study. ☐
- The information you have submitted will be published as a report; please tick this box if you would like to receive a copy. ☐

- I agree to take part in the above project. ☐
- I agree that the research team may use my data for future research and understand that in such cases, as with this project, data would not be identifiable in any report. ☐
- I agree to be contacted in the future by members of the hoarding research team at King's College London who would like to invite me to participate in follow up studies to this project, or in future studies of a similar nature. ☐

Participant's Statement:

I _____

agree that the research project named above has been explained to me to my satisfaction and I agree to take part in the study. I have read both the notes written above and the Information Sheet about the project, and understand what the research study involves.

Signed **Date**

Investigator's Statement:

I _____Claire Thompson_____confirm that I have carefully explained the nature, demands and any foreseeable risks (where applicable) of the proposed research to the participant.

Signed **Date**

INFORMATION SHEET FOR PARTICIPANTS *(PART 1)*

REC Reference Number **PNM/13/14-28**

The Experience of Caring in Hoarding (ECHO) – Study B

Version: 2

We would like to invite you to participate in this postgraduate research project. You should only participate if you want to; choosing not to take part will not disadvantage you in any way. Before you decide whether you want to take part, it is important for you to understand why the research is being done and what your participation will involve. Please take time to read the following information carefully and discuss it with others if you wish. Please ask us if there is anything that is not clear or if you would like more information.

What is the purpose of the study?

This study is an 'add on' to the study you have already participated in (Study A). It is part of a research project which aims to improve our understanding of the experiences faced by relatives (carers) of an individual who hoards and develop interventions to support them.

This part of the study involves piloting a new group intervention for relatives of people who hoard. We know that having a relative who hoards can be an upsetting and stressful experience, and that family members often feel frustrated and alone. We already know a little about some of the things that can be done to help relatives. We want to run a group to support these family members, and to give them an opportunity to meet and discuss hoarding with other people with similar experiences.

Why have I been chosen?

We are inviting 8 relatives of people who hoard to take part in the research who will be able to travel to South London for the groups.

Do I have to take part?

No. It is entirely up to you to decide whether or not to take part. If you decide to take part, you are free to withdraw from the research *at any time* and *without giving a reason*. Your decision about this will not affect the current or future involvement you may have with the research team

or health professionals or any care that your relative might currently receive or access in the future.

What will taking part involve for me?

- Participating will involve attendance at six group meetings over the course of six weeks from **…….**. Each group will take place on a Monday and last 2 hours. If you are happy to take part and the research team have answered any questions you have, you will be given this information sheet to keep and asked to sign a consent form.
- A brief outline of what you can expect from the groups is below:

Group meeting 1: In the first meeting members of the group will be given an opportunity to meet each other and share their experiences.

Group meetings 2-3: In these meetings the group will learn more about hoarding – including proposed causes and treatments.

Group meeting 4-5: In these meeting we will look at practical problem solving of common difficulties faced by relatives of people who hoard.

Group meeting 6: In the final meeting we will address coping strategies and how to look after your own well being, including relaxation and sleep.

Follow up group: There will be one more group meeting approximately a month after the group finished. The main purpose of this meeting will be to report any progress and trouble shoot difficulties encountered.

- After the groups are finished, we will ask you repeat some of the questionnaires you have filled in before for the first part of this study (Study A). You will already have an information sheet and consent form from Study A. After the follow up group we will ask you to repeat some of these again.

- We will reimburse your travel expenses of up to £10 per week of the group.

- If you agree to take part you will be asked whether you are happy to be contacted about participation in future studies. Your participation in this study will not be affected should you choose not to be re-contacted.

- You may withdraw from the research project at any time and can withdraw your information from the project up to one month after taking part

What are the possible benefits of taking part?

It is hoped that the group will give you the opportunity to meet other relatives of people who hoard and to share your experiences with each other. The group is also designed to give you information about hoarding and to discuss helpful strategies of how to cope and manage the emotional impact hoarding can have.

What are the possible disadvantages of taking part?

It is not expected that participation in the study has any risks. However, talking about your experiences of supporting a relative with hoarding difficulties might be upsetting for some people. If you would like to talk about this, please speak to the researcher on the contact details below.

If you have any questions or require more information about this study, please contact the researcher using the following contact details: Claire Thompson, Clinical Psychologist in Training, ASB, Institute of Psychiatry, 4 Windsor Walk, London, SE5 8AZ. Email: Claire.c.thompson@kcl.ac.uk, Tel: 0207 848 0733

If the study has harmed you in any way, please contact: Dr Juliana Onwumere, Department of Psychology, Henry Wellcome Building, Institute of Psychiatry, 16 De Crespigny Park, London, SE5 8AF Juliana.1.Onwumere@kcl.ac.uk, Tel: 020 7848 0197

Part 2

Will my taking part in the study be kept confidential?

All the information recorded will be strictly confidential and kept in accordance with the Data Protection Act 1998, and used only by clinicians and researchers working within the research team. Data from the study regarding you will be stored anonymously. We would only ever break this confidentiality in the case that you revealed something which caused us to be concerned for your safety or the safety of others, in which case we would inform you first and may need to contact appropriate services.

The questionnaires you complete will be kept in a locked cabinet at the research institution and only named researchers will have access to them. Your name will be kept separately with the number on a database and on paper so that we can identify your data in the future if we need to, for example, if you decide you no longer want to be part of the study. Your details will be kept for up to 4 years, and then will be confidentially

destroyed. We will keep a completely anonymised copy of the database indefinitely, from which you will not be able to be identified at all.

What will happen to the results of the study?

When the study is completed, we intend to publish the results in order to help other people working with family members of people who hoard, but your information would be completely confidential and you would not be named in the paper. We will also write to all participants and inform you of the results and offer you a copy of the report if you would like it.

Who has reviewed this study?

This study has been reviewed by Psychiatry, Nursing and Midwifery (PNM) Research Ethics Subcommittee (RESC) at King's College London, REC reference number: PNM/13/14-28

You will receive a copy of this information sheet and the signed consent section if you decide to participate.

Thank you for considering taking part in our study.

CONSENT FORM FOR PARTICIPANTS IN RESEARCH STUDIES

Please complete this form after you have read the Information Sheet and/or listened to an explanation about the research.

Title of Study: _____ECHO study B_____

King's College Research Ethics Committee Ref:___ PNM/13/14-28 ___

Thank you for considering taking part in this research. The person organising the research must explain the project to you before you agree to take part. If you have any questions arising from the Information Sheet or explanation already given to you, please ask the researcher before you decide whether to join in. You will be given a copy of this Consent Form to keep and refer to at any time.

- I confirm that I have read the information sheet dated for the above study. I have had the opportunity to consider the information and ask questions. ☐

- I understand my participation is voluntary, and that if I decide at any time during the research that I no longer wish to participate in this project, I can notify the researchers involved and withdraw from it immediately without giving any reason. ☐

- I consent to the processing of my personal information for the purposes explained to me. I understand that such information will be handled in accordance with the terms of the UK Data Protection Act 1998. ☐

- I understand that confidentiality and anonymity will be maintained and it will not be possible to identify me in any publications. ☐

- I consent to my interview being audio recorded and understand this will be deleted no later than 1 year after data has been entered. ☐

- I understand that I may withdraw my data from the study up to one month after I take part. Date of withdrawal _____ ☐

- I agree to respect the confidentiality of others contribtuions during group sessions (i.e. not revealing names or identifying details of other peoples experiences) ☐

- I agree to take part in the above project ☐

- I agree to be contacted in the future by members of the hoarding research team at King's College London who would like to invite me to participate in follow up studies to this project, or in future studies of a similar nature. ☐

- I agree that the research team may use my data for future research and understand that in such cases, as with this project, data would not be identifiable in any report. ☐

Participant's Statement:

I _____agree that the research project named above has been explained to me to my satisfaction and I agree to take part in the study. I have read both the notes written above and the Information Sheet about the project, and understand what the research study involves.

Signed **Date**

Investigator's Statement:

I _____Claire Thompson_____confirm that I have carefully explained the nature, demands and any foreseeable risks (where applicable) of the proposed research to the participant.

Signed **Date**

Appendix 4: Phase 2 group protocol

Week 1: Emotional processing: Opportunities to tell their story, addressing common emotional issues faced by relatives of people who hoard such as loss, guilt, and frustration. Goal: Normalisation and cognitive reappraisal in the context of severe problems.

1. Welcome to the group and introductions
2. Overview of the rest of the programme
3. Set group rules
4. Discussion of what it is like having a relative with a hoarding problem and sharing stories

Week 2: Understanding HD: Causes and symptoms. Goal: Improve carer attributions about relative and relationship quality; improve style of interaction with relative; reduce pressure on carer to clear house or make relative clear house, address concerns about carer developing the condition.

1. History of Hoarding
2. Definition of Hoarding
3. Main features of Hoarding
4. Who is affected
5. Reasons why people hoard
6. Reported causes

Week 3: Understanding HD: treatments and how you can help. Goal: As per week 2.

1. Discussion of treatments for hoarding
2. Difficulties and challenges in treatment
3. What can the carer do to help
4. Helpful Do's and Don'ts
5. Signposting to useful resources

Week 4-5: Problem-solving common difficulties faced by caregivers of hoarders. Goal: Optimise adaptive coping with specific problems; maintaining a relationship in the context of emotional reactions towards relative, cognitive reappraisal of problems, and helpful reactions.

1. Introduction to purpose of problem solving sessions
2. Brainstorm of problems faced by relatives of someone who hoards and group select up to 6 to cover over 2 weeks. Suggested problems included: Risk and safety, Dealing with irritability, Keeping your space – boundaries, Relationship with the person, Talking to them about the hoarding, Explaining hoarding to others, Fears of becoming a hoarder, Practical tips
3. Problem solving

Week 6: Promoting carer wellbeing. Goal: Improve carer strategies to look after themselves in terms of physical and mental wellbeing, e.g. sleep, relaxation, support networks; distancing from immediate problems, sense of perspective; impact on other relationships.

1. Carer stress
2. Strategies for managing stress
3. Setting goals and making personalised plan
4. Review of all group

Appendix 5: Measures

5.1 Carer Demographics sheet

Date: _____

Participant ID:		
DOB:	Age:	Gender:

Ethnicity

1st language: _____

Choose one option that best describes your ethnic group or background:

White

1. English / Welsh / Scottish / Northern Irish / British
2. Irish
3. Gypsy or Irish Traveller
4. Any other White background, please describe_____

Mixed / Multiple ethnic groups

5. White and Black Caribbean
6. White and Black African
7. White and Asian
8. Any other Mixed / Multiple ethnic background, please describe _____

Asian / Asian British

9. Indian
10. Pakistani
11. Bangladeshi
12. Chinese
13. Any other Asian background, please describe _____

Black / African / Caribbean / Black British

14. African
15. Caribbean
16. Any other Black / African / Caribbean background, please describe _____

Other ethnic group

17. Arab
18. Any other ethnic group, please describe_____

Marital Status

1 = *single* 2 = *married* 3 = *cohabiting (living with partner)*	4 = *divorced/separated* 5 = *widowed*	6 = *other (specify)*_____

Employment Status

1 = *employed full-time* 2 = *employed part-time* 3 = *voluntary employment*	4 = *unemployed* 5 = *housewife/husband* 6 = *student*	7 = *retired* 8 = *other (specify)*_____

How many days have you taken off work in the last 12 months to help the person who hoards?

_____

Education

1 = *primary education* 2 = *GCSE (O-level), 16yrs* 3 = *A-level, 18yrs*	4 = *undergraduate degree* 5 = *post graduate degree*	6 = *other (specify)*_____

What is your relationship to the person who hoards? They are my:

1 = *daughter* 2 = *son*	3 = *sister* 4 = *brother*	5 = *partner* 6 = *mother*	7 = *father* 8 = *friend*	9 = *other (specify)*_____

Age of the person who hoards: _____

Are you living with the person who hoards? Yes ☐ No ☐

How many hours of weekly contact do you have with the person who hoards: Hrs in contact each week _____

Is the person who hoards already seen by a mental health team: Yes ☐ N ☐

Do you provide care for anyone else: Yes ☐ *(specify below)* No ☐

1 = *daughter* 2 = *son*	3 = *sister* 4 = *brother*	5 = *partner* 6 = *mother*	7 = *father* 8 = *friend*	9 = *other (specify)*_____

In general, do you feel that you have someone that you can confide in? (talk to)
1 = *yes* 2 = *no*

5.2 The Structured Interview for Hoarding Disorder (SIHD; Nordsletten et al., 2013)

Permissions: The authors hold the copyright but the scale is free to use by researchers and clinicians who have an interest in hoarding disorder.

Citation: If you use the instrument, please cite it appropriately as follows: Nordsletten, A.E., Fernández de la Cruz, L., Pertusa, A., Reichenberg, A., Hatch, S.L., & Mataix-Cols, D. (2013). The Structured Interview for Hoarding Disorder (SIHD): Development, usage and further validation. *Journal of Obsessive Compulsive and Related Disorders*, 2(3), 346-350.

Correspondence: For any correspondence regarding the SIHD, please contact Dr Pertusa (alberto.pertusa@kcl.ac.uk) or Professor Mataix-Cols (david.mataix.cols@ki.se).

Instructions for the rater

The questions contained in this interview relate to each of the six criteria needed to evaluate the presence of hoarding disorder and its two specifiers. These questions appear in bold print and should be asked during the course of the interview, while the text in italics is present only to assist the rater. For a diagnosis of hoarding disorder all six criteria must be endorsed. If any of the criteria are not met, the diagnosis can be ruled out. The specifiers are only relevant for individuals endorsing all diagnostic criteria.

It is important to carefully distinguish hoarding disorder from non-pathological collecting, as well as from the general medical and DSM-5 conditions that may result in the accumulation of possessions (e.g., brain injury, obsessive-compulsive disorder, autism spectrum disorder, etc.). Therefore, this interview should ideally be used as a complement to a more comprehensive assessment of the patient's medical history and psychopathology. If in doubt about the endorsement of a specific criterion, the rater should complete the interview and consider all available information before rendering a diagnosis. Special sections are provided at the end of this document to assist with some of the most common differential diagnoses.

Ideally, the interview should be conducted directly with the sufferer and in the

person's home. If the individual of interest is not available or refuses to be interviewed, this interview may be administered to a reliable informant. This approach may also be employed for cases presenting with poor or absent insight, where the subjects responses may significantly conflict with the reality of the hoarding behavior. In cases where there is a strong clinical suspicion of HD (e.g. based on familial or legal reports), paired with poor insight on the part of the hoarding individual, the interviewer should use their clinical judgment in determining the relevance of each criterion.

If a home visit is not possible, photographs of the person's home environment may be helpful to assess the presence of clinically significant clutter (Criterion C). The presence of clutter may also be quantified with other available instruments such as the Clutter Image Rating Scale[5]. On the Clutter Image Rating Scale, a room score greater than 4 is usually indicative of clinically significant clutter, however this is only for guidance and all available information needs to be taken into account.

CRITERION A

Persistent difficulty discarding or parting with possessions, regardless of their actual value.

Does your relative experience difficulty discarding or parting with possessions? *This may include throwing away, selling, giving away, recycling, etc.*

☐ YES → *go to* ***next box***

☐ NO → *hoarding disorder not present*

How long has your relative had this problem for? _____ months/years.

[5] Frost, R.O., Steketee, G., Tolin, D.F., Renaud, S. (2008). Development and validation of the Clutter Image Rating. *Journal of Psychopathology and Behavioral Assessment, 30*(3):193-203.

If hoarding is a persistent problem that has been present for a long period of time → ***Criterion A is present*** *→ go to* ***next box***

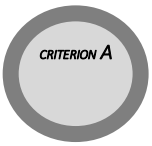
If hoarding has been present for a relatively short period of time (i.e., only a few weeks or months), inquire about temporary factors that may account for the difficulties discarding (e.g., recent inheritance of a large number of possessions, moving to a different home). If the hoarding behavior can be entirely explained by these circumstances → <u>*hoarding disorder not present*</u>

What items does your relative find it most difficult to discard? *Please list items below* (*both valuable and worthless items should be taken into account for the diagnosis).*

_____

_____

_____



If ***CRITERION A*** *is present, place a check in the circle and go to* ***CRITERION B***

CRITERION B

This difficulty is due to a perceived need to save items and to distress associated with discarding them.

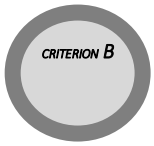
Does your relative intentionally keep these items (are they important/ useful for you)?

Does your relative generally feel distressed or upset when discarding possessions?

These questions are intended to evaluate whether the accumulation of objects is intentional/active and whether the discarding process causes distress (or would cause distress, in cases where discarding is entirely avoided). Where the accumulation is due to passive accumulation, or where the discarding process does not cause distress, the hoarding may be subclinical or attributable to an alternative psychopathology.

☐ If YES to both of the above questions → ***CRITERION B is present***

☐ If NO to any of the above questions → *hoarding disorder not present*



*If **CRITERION B** is present, place a check in the circle and go to **CRITERION C***

CRITERION C

The difficulty discarding possessions results in the accumulation of possessions that congest and clutter active living areas and substantially compromises their intended use. If living areas are uncluttered, it is only because of the interventions of third parties (e.g., family members, cleaners, authorities).

Does your relative have a large number of possessions which congest and clutter the main rooms in your home? *Note that "clutter" refers to the presence of a large number of items that are lying about in a disorganized way. The question refers to the key living spaces such as bedrooms, kitchen, or living room. Here exclude garages, attics, lofts, basements, and other areas that may commonly be cluttered in the homes of non-hoarding individuals.*

To meet Criterion C, active living spaces that are necessary for everyday life must be cluttered to the extent that their use is substantially compromised. If unclear, ask about the level of obstruction for particular rooms or domestic activities:

Because of the clutter or number of possessions, how difficult is it for you or your relative to use the rooms in your home?

- *Kitchen (sink, fridge, worktop, etc.):*

_____

- *Bathroom (sink, toilet, shower/bathtub, etc.):*

_____

- *Bedroom (bed, wardrobe, drawers, etc.):*

_____

- *Living room (sofa, chairs, table, floor, etc.):*

_____

- *Other (halls/corridors/stairs; difficult to walk through due to piles of items):*

_____

_____

☐ YES → ***CRITERION C is present***

☐ NO → *go to the* ***next box***

Have other people (such as family members or local authorities) helped your relative remove (or forcibly removed) some of your possessions? If so, how cluttered was their house/room before their intervention? *Explore to what extent the living spaces are currently clutter-free because of the intervention of other people. If this is the case, the criterion can be endorsed in the absence of significant clutter.*

☐ YES → ***CRITERION C is present***

☐ NO → *hoarding disorder not present*

If ***CRITERION C*** *is present, place a check in the circle and go to* ***CRITERION D***

CRITERION C

CRITERION D

The hoarding causes clinically significant distress or impairment in social, occupational, or other important areas of functioning (including maintaining a safe environment for self and others).

Do the difficulties discarding or the clutter cause your relative distress? *Note that some individuals with poor insight may not acknowledge being distressed, though any attempts to discard possessions by third parties will result in distress or anger.*

Do the difficulties discarding or the clutter interfere with their family life, friendships, or ability to perform well at home or work? *Note that the impairment may only be apparent to those around an individual with poor insight.*

☐ If YES to one or both of the above questions → ***CRITERION D is present***

☐ If NO to both questions → *hoarding disorder not present*

CRITERION D

*If **CRITERION D** is present, place a check in the circle and go to **CRITERION E***

CRITERION E

The hoarding is not attributable to another medical condition (e.g., brain injury, cerebrovascular disease, Prader-Willi syndrome).

Does your relative have any general medical conditions, a history of head injury or cerebrovascular disease? *Review past medical history for neurological disorders and inquire about history of severe head trauma. Some relevant conditions include traumatic brain injury, surgical resection for the treatment of a tumor or seizure control, cerebrovascular disease, infections of the central nervous system (e.g., herpes simplex encephalitis), or neurogenetic conditions such as Prader-Willi syndrome. If appropriate and available, additional investigations (e.g., EEG, CT, MRI, neuropsychological assessment) may be useful to help confirm the presence of brain damage.*

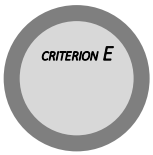
☐ YES → *go to* ***next question***

☐ NO → ***CRITERION E is present***

Did your relative have difficulties with discarding/clutter before they became ill? *Try to establish whether there is a clear temporal link between the medical condition and the onset of the hoarding behavior.*

☐ YES → ***CRITERION E is present***

☐ NO →*if hoarding clearly preceded by a general medical condition →<u>hoarding disorder not present</u>*



*If **CRITERION E** is present, place a check in the circle and go to **CRITERION F***

CRITERION F

The hoarding is not better explained by the symptoms of another mental disorder (e.g., obsessions in obsessive-compulsive disorder, decreased energy in major depressive disorder, delusions in schizophrenia or another psychotic disorder, cognitive deficits in major neurocognitive disorder, restricted interests in autism spectrum disorder).

Ideally this interview should be administered in the context of a full psychopathological assessment. If this is not available, ask the interviewee or informant about current or past psychiatric diagnoses. Note current and lifetime mental disorders here:

_____

_____

_____

_____

The presence of another mental disorder does not preclude the diagnosis of hoarding disorder. However, hoarding disorder is not diagnosed if the symptoms are judged to be secondary to or a direct consequence of another mental disorder, such as:

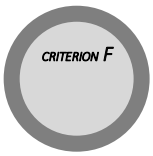
- *obsessions or compulsions in obsessive-compulsive disorder*
- *special or circumscribed interests in Autism Spectrum Disorder or intellectual disability*
- *decreased energy, psychomotor retardation or fatigue in Major Depressive Disorder*
- *delusions or negative symptoms in Schizophrenia Spectrum or other Psychotic Disorder*

- *cognitive deficits in a Neurocognitive disorder such as frontotemporal lobar degeneration or*

  Alzheimer's Disease

If another mental disorder is present, it is useful to establish the temporal relation with the onset of hoarding symptoms.

PLEASE SEE APPENDIX FOR FURTHER GUIDANCE ON THE DIFFERENTIAL DIAGNOSIS WITH OBSESSIVE-COMPULSIVE DISORDER AND AUTISM SPECTRUM DISORDER.



If **CRITERION F** *is present, place a check in the circle*

If all six criteria are met, the diagnosis of **hoarding disorder** *should be coded.*

If hoarding disorder is present, please place a check mark in the circle.

SPECIFIERS

If hoarding disorder has been diagnosed, assess the presence of Excessive Acquisition and determine the Degree of Insight.

EXCESSIVE ACQUISITION SPECIFIER

If the difficulty discarding possessions is accompanied by excessive acquisition of items that are not needed or for which there is no available space.

Does your relative often acquire free items that you don't need or for which you don't have available space at home?

☐ YES

☐ NO

Does your relative often buy items that you don't need, you can't afford, or for which you don't have available space at home?

☐ YES

☐ NO

Does your relative sometimes steal things that you don't need, you can't afford, or for which you don't have available space at home?

☐ YES

☐ NO

If YES *to* ***any*** *of the above 3 questions,* With Excessive Acquisition *should be coded.*

Please place a check mark in the circle.

INSIGHT SPECIFIER

With good or fair insight: *The individual recognizes that hoarding-related beliefs and behaviors (pertaining to difficulty discarding items, clutter, or excessive acquisition) are problematic.*

With poor insight: *The individual is mostly convinced that hoarding-related beliefs and behaviors (pertaining to difficulty discarding items, clutter, or excessive acquisition) are not problematic despite evidence to the contrary.*

With absent or delusional insight: *The individual is completely convinced that hoarding-related beliefs and behaviors (pertaining to difficulty discarding items, clutter, or excessive acquisition) are not problematic despite evidence to the contrary.*

To what extent do you think that your relative's saving behavior (including your difficulties discarding, the resulting clutter and the excessive acquisition) is problematic? *If in doubt, refer back to information provided by the subject during the interview. If a reliable informant is present, check for discrepancies between the subject's and the informant's report and assess degree of insight accordingly.*

☐ Good/Fair insight

☐ Poor insight

☐ Absent/Delusional insight

RISK ASSESSMENT

This section helps the rater document any possible risks associated with problematic hoarding behavior. Please check whether the following are present:

Fire hazard

☐ *Are there flammable materials near a heat source?*

☐ *Are there electrical hazards?*

Blocked exits

☐ *Is the door that allows entry and exit to the house clear?*

☐ *Are there additional doors within the property that are blocked?*

Risk of falling

☐ *Is there a lack of clear pathways, impeding movement throughout the property?*

☐ *Is it necessary to climb piles of objects in order to move between rooms or access*

objects?

Insects, infestations

☐ *Is there any evidence of insects (visible individuals, swarms, cobwebs, droppings)?*

☐ *Are there any rodents or other infestations present?*

Unhygienic conditions

☐ *Is there human or animal waste/vomit in the property?*

☐ *Is there moldy or rotten food or dirty food containers in the kitchen or other areas of the property?*

☐ *Is the sink, washbasin, bathroom, shower or bathtub clogged or notably dirty?*

☐ *Is there standing water anywhere in the property (sink, tub, basement, other)?*

☐ *Does the property emit a strong odor?*

Neglect of children, elder, or disabled people

☐ *If there are children, elders, or disabled people present, is there sufficient space to permit routine care and activities (e.g., a functioning kitchen, a place to eat meals, access to a shower or bathtub)?*

☐ *If there are children present, is there sufficient space for them to sleep, play, or do school homework?*

Animal hoarding

☐ *Are there starving, neglected, or maltreated animals on the premises?*

Additional notes *(please write any additional information that may be useful for risk assessment)*

_____

_____

_____

APPENDIX: DIFFERENTIAL DIAGNOSIS ASSISTANT

HOARDING AS A SYMPTOM OF OBESESSIVE-COMPULSIVE DISORDER

This section will assist the rater in assessing whether the hoarding behavior is better conceptualized as a symptom of obsessive-compulsive disorder (OCD). First, establish whether OCD is present (independently of the hoarding). If there is an established diagnosis of OCD, then ask the following questions:

Are your discarding difficulties caused by a specific obsession or fear?

☐ YES (more likely in OCD)

☐ NO

If hoarding is mainly driven by prototypical obsessions → <u>hoarding disorder probably not present</u> (hoarding likely to be a symptom of OCD)

Some examples of obsessions include:

- *not discarding for fear of contaminating self or others*
- *superstitious thoughts about discarding, e.g., fear of something bad happening to a loved one if certain items are discarded*
- *intense feelings of incompleteness*
- *saving to maintain a record of all life experiences*

Is it difficult for you to discard things because this triggers endless rituals (e.g., washing or checking rituals)?

☐ YES (more likely in OCD)

☐ NO

If hoarding is the result of persistent avoidance of onerous compulsions → <u>hoarding disorder probably not present</u> (hoarding likely to be a symptom of OCD)

Do you enjoy/find it comforting to acquire possessions and being around them?

☐ YES

☐ NO (more likely in OCD)

Are you emotionally attached to most of the items you save?

☐ YES

☐ NO (more likely in OCD)

Do you save items mainly because they are valuable/beautiful or they may come in handy in the future?

☐ YES

☐ NO (more likely in OCD)

Do you keep body products (feces, urine, nails, hair, used diapers) or rotten food?

☐ YES (more likely in OCD)

☐ NO

Individuals with hoarding disorder are more likely to report that their hoarding behavior is pleasurable/comforting, that they are emotionally attached to their saved objects, or that they save due to a belief that their items will prove handy in the future. The retention of body products or rotten food is, conversely, more often seen in OCD.

REMEMBER *that both OCD and hoarding disorder may be diagnosed at the same time when severe hoarding symptoms appear concurrently with other typical symptoms of OCD but are judged to be independent from these symptoms. In case of diagnostic uncertainty, we recommend diagnosing OCD only.*

HOARDING AS A SYMPTOM OF AUTISM SPECTRUM DISORDER

This section will assist the rater in assessing whether the hoarding behavior is better conceptualized as a symptom of Autism Spectrum Disorder (ASD). First, establish whether ASD is present (independently of the hoarding). If there is an established diagnosis of ASD, then ask the following questions:

Are the objects you save generally confined to a single, specific (circumscribed) area of interest?

A circumscribed interest, as seen in ASD, is typified by an intense interest in a specific, narrow, and often unusual topic. These interests may result in the accumulation of many similar objects, which are unified as exemplars of this area of interest. Individuals with hoarding disorder are more likely to accumulate a wide range of objects (e.g., not confined to a single area of interest, or unified by a

highly specific characteristic). A lack of organization is, furthermore, more typical in hoarding disorder.

☐ YES (more likely in ASD).

☐ NO

If yes *to the question above:* What is the area of interest?

_____

_____

_____

Do the objects you save largely share a particular, physical characteristic (e.g., material, texture or shape)?

☐ YES (more likely in ASD)

☐ NO

In ASD, the gathering of many like objects may signal an unusual, sensory preoccupation.

Examples of such preoccupations include intense fascinations with:

- *visual stimuli (e.g., shiny objects, blinking lights, the motion of liquid – such as the rotation of water being flushed)*
- *auditory stimuli (e.g., the sound of a vacuum cleaner)*
- *tactile stimuli (e.g., smooth surfaces)*

Do you enjoy organizing and classifying your possessions?

☐ YES (more likely in ASD)

☐ NO

If yes *to the question above:* Could you tell me a bit about your organizing system?

_____

_____

_____

A focus on uniformity and order with one's possessions is common to ASD. Unlike with OCD, in ASD this organization process should be egosyntonic and pleasurable.

If hoarding is the primarily the result of a circumscribed interest, sensory preoccupation or a desire to save/classify information → Hoarding disorder probably not present (hoarding likely to be a symptom of ASD)

***REMEMBER** that both ASD and Hoarding disorder may be diagnosed at the same time when severe hoarding symptoms appear concurrently with other typical symptoms of ASD but are judged to be independent from these symptoms. In case of diagnostic uncertainty, we recommend diagnosing ASD only.*

5.3 Hoarding Rating Scale-Interview (HRS-I; Tolin et al., 2010)

1. Because of the clutter or number of possessions, how difficult is it for your relative to use the rooms in their home?

0	1	2	3	4	5	6	7	8
Not at all difficult		Mild		Moderate		Severe		Extremely difficult

2. To what extent does your relative have difficulty discarding (or recycling, selling, giving away) ordinary things that other people would get rid of?

0	1	2	3	4	5	6	7	8
No difficulty		Mild		Moderate		Severe		Extreme difficulty

3. To what extent does your relative currently have a problem with collecting free things or buying more things than they need or can use or can afford?

0	1	2	3	4	5	6	7	8
No problem		Mild, occasionally (less than weekly) acquires items not needed, or acquires a few unneeded items		Moderate, regularly (once or twice weekly) acquires items not needed, or acquires some unneeded items		Severe, frequently (several times per week) acquires items not needed, or acquires many unneeded items		Extreme, very often (daily) acquires items not needed, or acquires large numbers of unneeded items

4. To what extent does your relative experience emotional distress because of clutter, difficulty discarding or problems with buying or acquiring things?

0	1	2	3	4	5	6	7	8
None/not at all		Mild		Moderate		Severe		Extreme

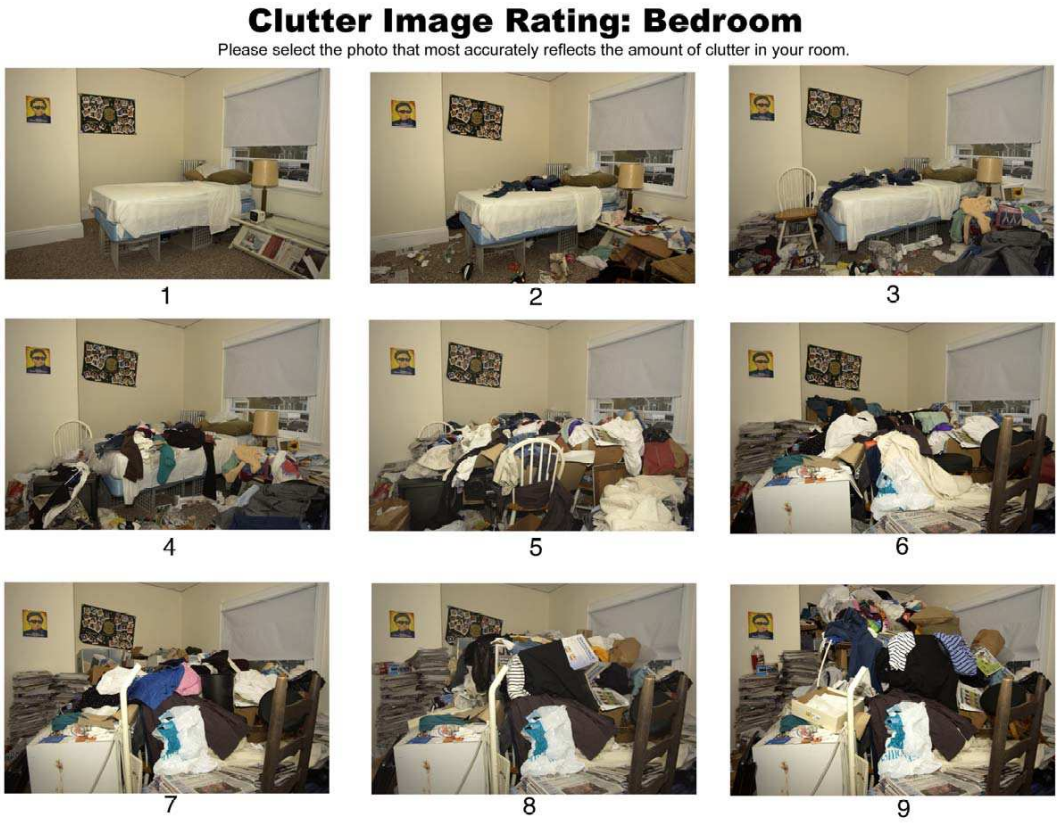
5. To what extent does your relative experience impairment in their life (daily routine, job/school, social activities, family activities, financial difficulties) because of clutter, difficulty discarding, or problems with buying or acquiring things?

0	1	2	3	4	5	6	7	8
None/not at all		Mild		Moderate		Severe		Extreme

5.4 Clutter Image Rating Scales (CIR; Frost et al., 2008)

Clutter Image rating scale: Bedroom

Your relatives Score:



Clutter Image rating scale: Kitchen

Your relatives Score:

Clutter Image rating scale: Living Room

Your relatives Score:

5.5 The Family Impact Scale for Hoarding (FISH; Nordsletten et al., 2014)

This scale is intended for use by relatives of individuals with hoarding problems. Where relevant, the scale may also be completed by unrelated persons who are living with or caring for someone with hoarding difficulties.

Section 1: Family Accommodation

In this section, we will be asking you about the extent to which you modify your behavior in order to prevent your relative's distress or avoid conflict with your relative. When responding, please indicate the extent to which you would agree with each statement over the PAST MONTH.

	Strongly Disagree	Somewhat Disagree	Somewhat Agree	Strongly Agree
1. I avoid discarding my relative's possessions due to the distress it will cause him/her.	0	1	2	3
2. I avoid discarding my own possessions due to the distress it will cause my relative.	0	1	2	3
3. I avoid discussing the discarding of items with my relative due to concerns that he/she will become angry or distressed.	0	1	2	3
4. I throw away belongings, tidy or clean in secret to avoid causing my relative irritation or distress.	0	1	2	3
5. I bring or buy my relative extra/unnecessary items because he/she asks me to do so.	0	1	2	3
6. I provide extra storage space for my relative's possessions (e.g., stored objects in my own bedroom, or home, rented storage space on my relative's behalf).	0	1	2	3
7. I shop on behalf of my relative in an attempt to limit his/her purchases.	0	1	2	3

8. Please tell us about other ways in which you have modified your life to ensure that your relative does not become distressed or upset:

Section 2: Burden

In this section we will ask you about the extent to which your relative's hoarding problem has had an impact on your OWN life. As with the previous section, please indicate the extent to which you would agree with each statement over the PAST MONTH.

	Strongly Disagree	Somewhat Disagree	Somewhat Agree	Strongly Agree
1. Due to the hoarding, I limit visits to the home of my relative, minimize the time spent at their home or arrange to meet with them elsewhere. (For those living with the hoarding individual, instead consider the following statement: Due to the hoarding, I try to minimize the amount of time I spend at home.)	0	1	2	3
2. Due to the hoarding, I am unable to use/have difficulty using the rooms in my relative's/our home for their intended purpose (e.g. have a family meal, watch TV, play with the children).	0	1	2	3
3. I have had to move out of my home due to my relative's hoarding.	0	1	2	3
4. I have had to buy/rent another property to live in due to my relative's hoarding.	0	1	2	3
5. I have modified my work/education schedule due to my relative's hoarding.	0	1	2	3
6. I have missed work/school due to my relative's hoarding.	0	1	2	3
7. I have modified my social life (e.g., not been able to bring friends home) due to my relative's hoarding.	0	1	2	3
8. I have modified my leisure activities (e.g., sports, hobbies) due to my relative's hoarding.	0	1	2	3
9. I have had loss of earnings due to my relative's hoarding.	0	1	2	3
10. I feel that the needs of my relative control my family life and activities.	0	1	2	3

11. Please tell us about other ways in which your relative's hoarding has had an impact on your life:

_____

_____

Permissions: The authors hold the copyright this scale, however the measure is free to use by researchers and clinicians who have an interest in hoarding.

Citation: If you use this instrument, please cite it appropriately as follows: Nordsletten, A.E., Fernández de la Cruz, L., Drury, H., Ajmi, S. & Mataix-Cols, D. The Family Impact Scale for Hoarding Disorder (FISH), (*to be submitted for publication)*

Correspondence: For any inquiries regarding the FISH, please contact Mrs. Nordsletten (ashley.nordsletten@gmail.com) or Professor Mataix-Cols (david.mataix-cols@kcl.ac.uk)

Scoring Instructions

Each closed-response item on the FISH is scored from 0 ("Strongly Disagree) to 3 ("Strongly Agree"). Scores may be aggregated over the entire measure to form a composite or, if preferred, by sub-scale to quantify a particulate element of family impact (e.g. "Burden").

When calculating a composite, scores will range between 0 and 45. Scores on the subscales will vary, with sums on "Family Accommodation" ranging between 0 and 18, while scores on "Burden" will range from 0 to 27. Regardless of the method chosen, increasing scores will always indicate increasing instance and degree of negative impacts.

As a guide for interpreting the severity of responses: initial application of this scale in a sample of individuals with hoarding relatives indicated a mean score of 20.52 (95% C.I. 17.58 – 22.45), while administration in a comparator group with healthy relatives produced a mean score of 6.73 (95% C.I. 4.41-9.05).

For further details, please refer to:

Nordsletten, A.E., Fernández de la Cruz, L., Drury, H., Ajmi, S. & Mataix-Cols, D. The Family Impact Scale for Hoarding Disorder (FISH), (*2014)*

5.6 Experience of Caregiving Inventory (Szmukler et al., 1996)

The following statements commonly apply to persons who care for relative or friends with a serious mental illness. We would like you to read each statement and decide how often it has applied to you over the **PAST ONE MONTH.**

If it has **never** happened or **rarely** happened you would CIRCLE the number 0 or 1. If it has happened **sometimes**, then you would CIRCLE the number 2. If it has happened **often** or seems to have happened **nearly always**, then you would CIRCLE the number 3 or 4.

It is important to note that there are no right or wrong answers. Also it is best not to spend too long on any one statement. Often your first reaction will usually provide the best answer. While there seem to be a lot of statements, you will find that it won't take more than a moment or so to answer each one.

During the past month *how often have you thought about:*

	NEVER	***RARELY***	***SOME-TIMES***	***OFTEN***	***NEARLY ALWAYS***
1. Your covering up their illness……..……	0	1	2	3	4
2. Feeling unable to tell anyone of the illness…...……..…...	0	1	2	3	4
3. Their difficulty looking after money...……..…	0	1	2	3	4
4. Having to support them……..……	0	1	2	3	4
5. What life they might have had……...……	0	1	2	3	4
6. Their risk of committing suicide…….....	0	1	2	3	4
7. I have learnt more about myself……..	0	1	2	3	4
8. I have contributed to others understanding of the illness….....	0	1	2	3	4
9. Being unable to do the things you want to do……..…..	0	1	2	3	4
10. How health professionals do not take you seriously……....	0	1	2	3	4
11. Their dependence on you……...…….….	0	1	2	3	4
12. Helping them to fill in the day……..…	0	1	2	3	4
13. I have contributed to their wellbeing……..……..……	0	1	2	3	4

14. That they make a valuable contribution to the household…...	0	1	2	3	4
15. The effect on your finances if they become more seriously ill	0	1	2	3	4
16. Dealing with psychiatrists…….. ……	0	1	2	3	4
17. Always having them at the back of your mind……..…	0	1	2	3	4
18. Whether you have done something to make them ill……..….	0	1	2	3	4
19. That they have shown strengths in coping with their illness…	0	1	2	3	4
20. I have become more confident in dealing with others……..	0	1	2	3	4
21. How family members do not understand your situation…..…..	0	1	2	3	4
22. That they are good company……..……..	0	1	2	3	4
23. I have become more understanding of others with problems..	0	1	2	3	4
24. How they think a lot about death……..	0	1	2	3	4
25. Their lost opportunities…… ……..	0	1	2	3	4
26. How to deal with mental health professionals……..…..	0	1	2	3	4
27. Feeling unable to have visitors at home……..……	0	1	2	3	4
28. How they get on with other family members……...…….	0	1	2	3	4
29. Backing them up when they run out of money……...….	0	1	2	3	4

	NEVER	RARELY	SOME-TIMES	OFTEN	NEARLY ALWAYS
1. How family members do not understand the illness……...…..	0	1	2	3	4
2. How they deliberately attempt to harm themselves…..……	0	1	2	3	4

3.	I have become closer to some of my family……	0	1	2	3	4
4.	I have become closer to friends……...	0	1	2	3	4
5.	I share some of their interests……..……..	0	1	2	3	4
6.	I feel useful in my relationship with them…….	0	1	2	3	4
7.	How health professionals do not understand your situation....	0	1	2	3	4
8.	Whether they will ever get well……..	0	1	2	3	4
9.	Feeling the stigma of having a mentally ill relative……	0	1	2	3	4
10.	How to explain their illness to others……	0	1	2	3	4
11.	Others leaving home because of the effect of their illness…..	0	1	2	3	4
12.	Setting them up in accommodation……..	0	1	2	3	4
13.	How to make complaints about their care……...	0	1	2	3	4
14.	I have met helpful people……...	0	1	2	3	4
15.	I have discovered strengths in myself…….....	0	1	2	3	4
16.	Feeling unable to leave them home alone……	0	1	2	3	4
17.	The effect of the illness on children in the family……...	0	1	2	3	4
18.	The illness causing a family breakup……...	0	1	2	3	4
19.	Them keeping bad company…….	0	1	2	3	4
20.	How their illness effects special family events……..…	0	1	2	3	4
21.	Finding out how hospitals or mental health services work…..	0	1	2	3	4
22.	Doctor's knowledge of the services available to families….....	0	1	2	3	4
23.	The difficulty getting information about their illness……...	0	1	2	3	4

During the past month *how often have you thought about her being:*

24. Moody……	0	1	2	3	4
25. Unpredictable……	0	1	2	3	4
26. Withdrawn……	0	1	2	3	4
27. Uncommunicative……	0	1	2	3	4
28. Not interested……	0	1	2	3	4
29. Slow at doing things……	0	1	2	3	4
30. Unreliable about doing things……	0	1	2	3	4
31. Indecisive……	0	1	2	3	4
32. Irritable……	0	1	2	3	4
33. Inconsiderate……	0	1	2	3	4
34. Behaving in a reckless way……	0	1	2	3	4
35. Suspicious……	0	1	2	3	4
36. Embarrassing in appearance……	0	1	2	3	4
37. Behaving in a strange way……	0	1	2	3	4

Thank you very much for answering these questions

5.7 Brief Illness Perceptions Questionnaire (Broadbent et al., 2006)

For the following questions, please circle the number that best corresponds to your views:

How much do you think your relative's illness affects his/her life?

0 1 2 3 4 5 6 7 8 9 10

no effects at all — severely affects his/her life

How long do you think your relative's illness will continue?

0 1 2 3 4 5 6 7 8 9 10

a very short time — forever

How much control do you feel your relative has over his/her illness?

0 1 2 3 4 5 6 7 8 9 10

absolutely no control — extreme amount of control

How much do you think your relative's treatment can help his/her illness?

0 1 2 3 4 5 6 7 8 9 10

not at all — extremely helpful

How much does your relative experience symptoms from his/her illness?

0 1 2 3 4 5 6 7 8 9 10

no symptoms at all — many severe symptoms

How concerned are you about your relative's illness?

0 1 2 3 4 5 6 7 8 9 10

not at all concerned — extremely

How well do you feel you understand your relative's illness?

0 1 2 3 4 5 6 7 8 9 10

don't understand at all — understand very clearly

How much does your relative's illness affect you emotionally? (eg. does it make you angry, scared, upset or depressed?)

0 1 2 3 4 5 6 7 8 9 10

not at all affected emotionally — extremely affected emotionally

Please list in rank-order the three most important factors that you believe caused <u>your</u> relative's <u>illness</u>. *The most important causes of my relative's illness are:-*

1. _____

2. _____

3. _____

5.8 Brief COPE (Carver, 1997)

These items deal with ways you've been coping with the stress in your life since your relative has been hoarding. There are many ways to try to deal with problems. These items ask what you've been doing to cope with this one. Obviously, different people deal with things in different ways, but I'm interested in how you've tried to deal with it. Each item says something about a particular way of coping. I want to know to what extent you've been doing what the item says, how much or how frequently. Don't answer on the basis of whether it seems to be working or not—just whether or not you're doing it. Use these response choices. Try to rate each item separately in your mind from the others. Make your answers as true FOR YOU as you can.

1 = I haven't been doing this at all
2 = I've been doing this a little bit
3 = I've been doing this a medium amount
4 = I've been doing this a lot

	Score 1-4 (please write in box)
1. I've been turning to work or other activities to take my mind off things.	
2. I've been concentrating my efforts on doing something about the situation I'm in.	
3. I've been saying to myself "this isn't real.".	
4. I've been using alcohol or other drugs to make myself feel better.	
5. I've been getting emotional support from others.	
6. I've been giving up trying to deal with it.	
7. I've been taking action to try to make the situation better.	
8. I've been refusing to believe that it has happened.	
9. I've been saying things to let my unpleasant feelings escape.	
10. I've been getting help and advice from other people.	

11. I've been using alcohol or other drugs to help me get through it.	
12. I've been trying to see it in a different light, to make it seem more positive.	
13. I've been criticizing myself.	
14. I've been trying to come up with a strategy about what to do.	
15. I've been getting comfort and understanding from someone.	
16. I've been giving up the attempt to cope.	
17. I've been looking for something good in what is happening.	
18. I've been making jokes about it.	
19. I've been doing something to think about it less, such as going to movies, watching TV, reading, daydreaming, sleeping, or shopping.	
20. I've been accepting the reality of the fact that it has happened.	
21. I've been expressing my negative feelings.	
22. I've been trying to find comfort in my religion or spiritual beliefs.	
23. I've been trying to get advice or help from other people about what to do.	
24. I've been learning to live with it.	
25. I've been thinking hard about what steps to take.	
26. I've been blaming myself for things that happened.	
27. I've been praying or meditating.	
28. I've been making fun of the situation	

Carver, C. S. (1997). You want to measure coping but your protocol's too long: Consider the Brief COPE. *International Journal of Behavioral Medicine*, 4, 92-100

Scales are computed as follows (with no reversals of coding):

Self-distraction, items 1 and 19
Active coping, items 2 and 7
Denial, items 3 and 8
Substance use, items 4 and 11
Use of emotional support, items 5 and 15
Use of instrumental support, items 10 and 23
Behavioural disengagement, items 6 and 16
Venting, items 9 and 21
Positive reframing, items 12 and 17
Planning, items 14 and 25
Humour, items 18 and 28
Acceptance, items 20 and 24
Religion, items 22 and 27
Self-blame, items 13 and 26

5.9 Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983)

Please circle one option for each question which best represents how you have been feeling over the last week. Do not spend too long on any one question, your first answer is often the most accurate.

A

I feel tense or 'wound up':

Most of the time 3

A lot of the time 2

From time to time, occasionally 1

Not at all 0

D

I still enjoy the things I used to enjoy:

Definitely as much 0

Not quite so much 1

Only a little 2

Hardly at all 3

A

I get a sort of frightened feeling as if something awful is about to happen:

Very definitely and quite badly 3

Yes, but not too badly 2

A little, but it doesn't worry me 1

Not at all 0

D

I can laugh and see the funny side of things:

As much as I always could 0

Not quite so much now 1

Definitely not so much now 2

Not at all

A

Worrying thoughts go through my mind:

A great deal of the time 3

A lot of the time 2

From time to time, but not too often 1

Only occasionally 0

D

I feel cheerful:

Not at all 3

Not often 2

Sometimes 1

Most of the time 0

A

I can sit at ease and feel relaxed:

Definitely 0

Usually 1

Not Often 2

Not at all

D

I feel as if I am slowed down:

Nearly all the time 3

Very often 2

Sometimes 1

Not at all 0

A

I get a sort of frightened feeling like 'butterflies' in the stomach:

Not at all 0

Occasionally 1

Quite Often 2

Very Often 3

D

I have lost interest in my appearance:

Definitely 3

I don't take as much care as I should 2

I may not take quite as much care 1

I take just as much care as ever 0

A

I feel restless as I have to be on the move:

Very much indeed 3

Quite a lot 2

Not very much 1

Not at all 0

D

I look forward with enjoyment to things:

As much as I ever did 0

Rather less than I used to 1

Definitely less than I used to 2

Hardly at all 3

A

I get sudden feelings of panic:

Very often indeed 3

Quite often 2

Not very often 1

Not at all 0

D

I can enjoy a good book or radio or TV program:

Often 0

Sometimes 1

Not often 2

Very seldom 3

Scoring (add the As = Anxiety. Add the Ds = Depression). The norms below will give you an idea of the level of Anxiety and Depression.

0-7 = Normal

8-10 = Borderline abnormal

11-21 = Abnormal

5.10 The Warwick-Edinburgh Mental Well-being Scale (WEMWBS; Tennant et al., 2007)

Below are some statements about feelings and thoughts.

Please tick the box that best describes your experience of each over the last 2 weeks

STATEMENTS	None of the time	Rarely	Some of the time	Often	All of the time
I've been feeling optimistic about the future	1	2	3	4	5
I've been feeling useful	1	2	3	4	5
I've been feeling relaxed	1	2	3	4	5
I've been feeling interested in other people	1	2	3	4	5
I've had energy to spare	1	2	3	4	5
I've been dealing with problems well	1	2	3	4	5
I've been thinking clearly	1	2	3	4	5
I've been feeling good about myself	1	2	3	4	5
I've been feeling close to other people	1	2	3	4	5
I've been feeling confident	1	2	3	4	5
I've been able to make up my own mind about things	1	2	3	4	5
I've been feeling loved	1	2	3	4	5
I've been interested in new things	1	2	3	4	5
I've been feeling cheerful	1	2	3	4	5

Warwick-Edinburgh Mental Well-Being Scale (WEMWBS)

Appendix 6: 'Knowledge about Hoarding Questonnaire' developed by the research team

Hoarding Questionnaire

(Thompson, Fernández de la Cruz, Mataix-Cols & Onwumere, 2014)

1. Hoarding is a type of :
    a. Mental health problem (1)
    b. Personality
    c. Problem of having low standards of living

2. Approximately what percentage of the population has Hoarding Disorder?
    a. Less than 1%
    b. 1.5% (1)
    c. 2-4%
    d. Up to 5%

3. What is the primary symptom of Hoarding Disorder?
    a. Laziness
    b. Difficulties discarding possessions (1)
    c. Wanting to recycle
    d. Emotional attachment to seemingly worthless possessions

4. What is the youngest age someone might start to show symptoms of hoarding?
    a. Early teens (12/13) (1)
    b. 20's
    c. 30's
    d. 40's

5. What are the reported reasons why some people are more likely to start hoarding?
    a. Perfectionism (needing everything to be 'just right')
    b. Difficulties with memory, attention and making decisions
    c.Suffering a traumatic life event (e.g. loss of a loved one)
    d. Genetics (i.e. can run in families)
    e. It could be any of the above (1)

6. What is the best way to help people with their hoarding difficulties?
    a. Throw things away without their knowledge (in secret)
    b. Ignore the hoarding
    c. Helping them making decisions about what to discard and what to keep (1)
    d. Convince them to move house in order to have more space available

7. What impact can hoarding have on family members of people who hoard?
    a. Embarrassment
    b. Fear of what will happen to their hoarding relative
    c. Reluctance to visit the persons home
    d. Loss of contact and relationship with the person who hoards
    e. Frustration and anger with their hoarding relative
    f. Any or all of the above (1)

Appendix 7: Table 12 - Means, standard deviations and t-test comparisons for group outcomes at pre, post and 1-month follow-up

				T-tests								
Variable	*Mean (SD)*			*Pre-post (n=11)*			*Pre-FU (n=8)*			*Post-FU (n=8)*		
	Pre-intervention (N=11)	*Post-intervention (N=11)*	*1-Month FU (N=8)*	*t=*	*p=*	*95% CI*	*t=*	*p=*	*95% CI*	*t=*	*p=*	*95% CI*
Wellbeing (WEMWBS)	29.18 (7.07)	33.55 (10.28)	45.25 (8.92)	-2.81	.018*	[-7.82, 0.91]	-5.05	.001*	[-23.13, -8.37]	-3.61	.009*	[-19.25, -4.00]
Distress (HADS)	13.64 (6.04)	13.91 (6.82)	13.25 (4.27)	-.20	.846	[-3.23, 2.78]	-.53	.610	[-6.11, 3.86]	-.43	.677	[-4.02, 2.78]
ECI negative	87.91 (30.64)	87.27 (36.92)	89.00 (8.45)	.12	.906	[-11.06, 12.33]	-.95	.373	[-32.24, 13.74]	-.63	.546	[-26.62, 15.37]
ECI positive	24.91 (10.41)	28.91 (10.00)	25.63 (9.27)	-2.97	.014*	[-7.00, -0.99]	-3.94	.006*	[-8.20, -2.05]	.31	.769	[-3.37, 4.37]
Avoidant coping (brief-COPE)	12.36 (2.73)	12.45 (3.05)	12.50 (2.27)	-.079	.938	[-2.65, 2.47]	.26	.802	[-2.01, 2.52]	.11	.913	[-2.50, 2.75]
Active coping	5.45 (2.02)	6 (1.41)	5.25 (1.67)	-1.20	.258	[-1.56, 0.47]	-.15	.882	[-2.04, 1.79]	1.26	.250	[-0.55, 1.80]

Emotional support	4.45 (1.75)	4.45 (1.57)	5.25 (1.49)	.00	1	[0.38, -0.85]	-1.39	.217	[-1.41, 0.46]	-3.00	.020*	[-1.34, 0.59]
Instrumental coping	5.09 (1.81)	5.27 (2.10)	5.12 (1.96)	-.36	.724	[-1.85-1.35]	-1.00	.351	[-1.26, 0.51]	-.16	.879	[-1.99, 1.74]
Reframing	3.91 (1.51)	4.36 (1.86)	3.37 (1.51)	-1.84	.096	[-1.01, 0.96]	-.23	.826	[-1.42, 1.71]	.31	.763	[-0.82, 1.07]
Planning	5.82 (1.25)	6.37 (1.27)	5.37	-1.34	.211	[-1.21, 0.30]	.00	1	[-1.79, 1.79]	1.36	.217	[-0.46, 1.71]
Humour	4.18 (1.47)	3.73 (1.42)	3.5 (1.31)	1.05	.320	[-0.51, 1.42]	.51	.626	[-0.91, 1.41]	-.68	.516	[-1.12, 0.62]
Acceptance	5.09 (1.58)	5.27 (1.68)	5.13 (1.36)	-.61	.553	[-0.84, 0.48]	-.63	.549	[-1.78, 1.03]	-.61	.563	[-1.22, 0.72]
Religion	3.10 (1.38)	3.36 (1.36)	3.50 (1.60)	-1.94	.082	[-0.59, 0.04]	-1.53	.170	[-1.27, 0.27]	-.61	.563	[-1.22, 0.72]

Appendix 8: Table 13 - Means and Standard Deviations on main outcome measures for phase 1: Adult children vs other relatives

Measure	*Adult Children - Mean (SD)*	*Other relatives - Mean (SD)*
ECI negative	78.86 (29.97)	103.71 (45.07)
ECI positive	19.64 (9.09)	24.71 (9.39)
B-IPQ total	56.25 (5.66)	58.47 (7.92)
Brief COPE avoidant	12.94 (2.98)	13.88 (4.79)
HADs total	14.31 (6.76)	16.06 (11.65)
% depressed	8.3%	11.8%
% anxious	36%	52.9%
% anxious or depressed	38.9%	52.9%
WEMWBS	30.14 (8.13)	29.12 (12.13)
% High EE	63.9%	35.3%
FISH total	37.5 (6.99)	40.82 (11.47)
FISH burden	21.89 (5.49)	25.06 (7.68)
FISH accommodation	15.61 (2.96)	15.76 (4.99)

Institute of Psychiatry, Psychology and Neuroscience

King's College London

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Service Related Project

Investigating appropriate cut-off scores on Rivermead Post-Concussion Symptoms Questionnaire (RPQ) in a sample of outpatients at the Lishman Brain Injury Unit.

Supervised by:

Dr Sebastian Potter

Dr Mary Summers

Lishman Brain Injury Unit, South London and Maudsley NHS Foundation Trust

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Abstract

The present study investigated the specificity and sensitivity of a widely used measure of post-concussion symptoms, the Rivermead Post-concussion Symptoms Questionnaire (RPQ) by comparing a group of patients from the Lishman Brain Injury Unit at the Maudsley Hospital in London, who had a traumatic brain injury at least 6 months previously and met ICD-10-DCR criteria for Post-concussion Syndrome, to a control group without traumatic brain injury. It also aimed to establish the optimal cut-off score for the RPQ and compare classification accuracy at existing and optimal cut-off points. Multiple versions of the RPQ derived from the literature were investigated using receiver-operating characteristic (ROC) curve analysis, and compared as to their ability to accurately distinguish PCS patients (n=61) from healthy controls (n=46). The RPQ was shown to have high classification accuracy, as did other suggested versions of the RPQ. Using the results of the ROC analysis, an optimal cut-off point of 16 on the RPQ was selected as having maximum sensitivity (97%) and specificity (87%) to distinguish from controls. Implications for practise at the Lishman Unit and other brain injury services are discussed.

1. Service context and Aims of the Project

The Lishman Brain Injury Unit at the Maudsley Hospital is a national, specialist service that sees up to 280 outpatients a year with neuropsychiatric sequelae in the context of acquired brain injuries. Many of these patients have suffered mild to moderate traumatic brain injury with enduring effects on their wellbeing and daily functioning and are seen for neuropsychological assessment and treatment. Routine questionnaires administered along with tests of cognitive function include the Hospital Anxiety and Depression Scale (HADS, Zigmond & Snaith, 1983) and the Rivermead Post-Concussion Symptoms Questionnaire (RPQ, King, Crawford, Wenden, Moss, & Wade, 1995), as it is known a significant proportion of people suffer from anxiety, depression and other adverse effects following even a mild traumatic brain injury. The psychometric qualities and predictive value of the HADS are well established, but post-concussion symptoms can be challenging to diagnose and formulate, as will be detailed in the following literature review. The accurate diagnosis of post-concussion symptoms is important within the service to ensure that patients receive appropriate treatment and formulation of problems which may affect them daily. Over-diagnosis may lead to iatrogenic effects such as excess disability. Likewise, under-diagnosis has negative implications both for the patient (e.g. lack of explanation for symptoms, feeling invalidated) and the service (e.g. underestimating the numbers of patients with the problem, lack of appropriate training and resource allocation). Therefore, this study aims to investigate the sensitivity, specificity and optimal cut-off score of the RPQ in order to efficiently identify post-concussion syndrome. This would help to improve patient quality of care within the service and more widely in other brain injury services.

2. Introduction

2.1 Traumatic Brain injury

Mild traumatic brain injury (MTBI) is the most common form of brain injury, making up approximately 90% of emergency hospital presentations (Kay & Teasdale, 2001). An MBTI is defined by the American Congress of Rehabilitation Medicine (ACRM, 1993): “A patient with mild traumatic brain injury is a person who has had a

traumatically induced physiological disruption of brain function, as manifested by at least one of the following:

1. Any period of loss of consciousness;

2. Any loss of memory for events immediately before or after the accident;

3. Any alteration in mental state at the time of the accident (e.g., feeling dazed, disoriented, or confused); and

4. Focal neurological deficits that may or may not be transient; but where the severity of the injury does not exceed the following:

- Loss of consciousness of approximately 30 minutes or less;
- after 30 minutes a Glasgow Coma Scale (GCS) of 13-15 and
- Posttraumatic amnesia (PTA) not greater than 24 hours."

2.2 Post-concussion syndrome

While most people suffer no adverse effects following MTBI, around half may experience some post-concussion symptoms. These can be divided into three main symptom clusters, i.e. cognitive (e.g. poor memory or concentration), emotional (e.g. irritability, depression, anxiety) and physical (e.g. headaches, dizziness, fatigue, visual disturbances, noise sensitivity) symptoms. For the majority of patients these symptoms resolve within a few months. However, for a "miserable minority", these symptoms can endure for months and even years after a TBI (Rohling et al., 2011; Ruff, Camenzuli, & Mueller, 1996). Where these symptoms endure for months after the TBI, a diagnosis of Post-Concussion Syndrome can be made (See Appendix 1 & 2 for ICD-10 and DSM-IV diagnostic criteria). Prevalence of PCS is estimated to be between 15-30% of people with MTBI (King, 2003; Wood, 2004). Neither DSM-IV nor ICD-10 criteria explicitly specify the minimal or maximal injury severities associated with PCS, but it is usually associated with mild traumatic brain injury.

PCS has been the subject of controversy for over 100 years (King, 2003). Firstly, the diagnostic criteria are not widely accepted, and may be of limited usefulness in identifying PCS. A study by Boake et al. (2005) showed that prevalence of PCS was about six times higher with ICD-10 than with DSM-IV criteria and that specificity of diagnosis was limited with regard to brain injury; patients could meet criteria even

without brain damage, e.g. having sustained head trauma but no TBI. The authors found that whereas the DSM-IV criteria were more stringent than ICD-10, the inclusion of cognitive impairment and clinical significance did not increase specificity (as again these are not limited to brain injury). Another study by Kashluba, Casey and Paniak (2006) found that the ICD-10 criteria has a 39% false positive rate, incorrectly classifying control participants as having PCS and at 3 months post-injury could not differentiate between control and TBI groups. The authors concluded that the classification accuracy of the ICD-10 criteria could only be described as 'fair'.

Secondly, the reason for the lack of specificity of diagnosis may be that many PCS symptoms are not specific to PCS and occur commonly in the general population (Chan, 2001; Iverson & Lange, 2003; Wang, Chan, & Deng, 2006) and other patient groups, such patients with chronic pain (Smith-Seemiller, Fow, Kant, & Franzen, 2003) and depression (Iverson, 2006). Herrmann and colleagues (2009) found that depressed patients with TBI scored higher on all items on the RPQ than non-depressed patients with TBI, and therefore PCS symptoms may overlap or be exaggerated by other disorders. This is important to be aware of, as several studies have shown that psychiatric comorbidity is common in PCS patients (Potter & Brown, 2012). Additionally, there appears to be a gender bias; according to a recent study by Styrke, Sojka, Björnstig, Bylund, & Stålnacke (2013) there are gender differences in PCS symptomatology, with women more likely to report PCS symptoms and meet diagnostic criteria for PCS and experience a higher level of disability.

Thirdly, there has been much debate about whether the causes of PCS are biological, psychological, or a combination of both. Most people make a full recovery from MTBI and show little evidence of prolonged cognitive difficulties or other symptoms; a meta-analysis by Rohling et al. (2011) found that MTBI had only a very small and non-significant effect size on neuropsychological functioning 3 months post-injury (d=0.07), although residual damage to the brain may be observed (e.g. Anderson, Heitger, & Macleod, 2006, see King, 2003, for a review). On the other hand, many psychological factors have been demonstrated to play a part in the persistence of PCS, such as pre-existing psychopathology, medico legal claims, stress, depression and the

role of expectations (Wood, 2007). Diathesis-stressor models (Lishman, 1988; Kay, 1993; Wood, 2007) attempt to reconcile these positions by involving a recognition of early direct effects of injury, but proposing that symptoms may be maintained through other mechanisms.

2.3 Treatment of Post-concussion syndrome

PCS symptoms are often treated with medications, most often non steroidal anti-inflammatory analgesics and antidepressants (Mittenberg, Canyock, Condit, & Patton, 2001). However, Mittenberg and Burton (1994) surveyed professional members of the Neuropsychological Society and National Academy of Neuropsychology and found the treatments professionals reported using most were education, support and reassurance. Treatment approaches may involve early prophylactic interventions, such as information leaflets (Mittenberg, Zielinski, & Fichera, 1993) or more comprehensive psychological treatment such as cognitive rehabilitation or CBT if symptoms persist.

The primary aims of psychological treatment of PCS are to normalise symptoms, minimise the vicious circle of stress and PCS symptoms maintaining each other, facilitate re-introduction of activities and prevent catastrophic misinterpretations of symptoms (King, 2003). CBT for PCS would also usually include anxiety management, coping strategies and cognitive restructuring (Potter & Brown, 2012; Tiersky et al., 2005). A systematic review by Sayegh, Sandford, & Carson (2010) found that CBT for PCS was an effective treatment, showing promising outcomes for mood and PCS symptoms in several trials. Potter and colleagues (Potter, Brown, & Fleminger, submitted) have recently completed an RCT of CBT for persistent PCS at the Lishman Brian Injury Unit with 46 patients completing 12 sessions of CBT focussed around PCS symptoms. Preliminary results suggest a positive impact of therapy on quality of life and symptoms.

2.4 Assessment of Post-concussion syndrome

As well as the ICD-10 and DSM-IV criteria for the diagnosis of PCS, a number of questionnaires and checklists also exist which are used in research and clinical practise

to identify the presence of PCS symptoms. The most widely cited measure is the Rivermead Post-Concussion Symptoms Questionnaire (RPQ, see Appendix 3, King, Crawford, Wenden, Moss, & Wade, 1995). The RPQ is a self-report measure of the severity of PCS symptoms. It consists of 16 items asking the patient whether post-concussion symptoms have been experienced over the previous 24 hours compared to before the brain injury, namely: headaches, fatigue, dizziness, nausea, noise sensitivity, sleep disturbance, irritability, depression frustration, poor memory, poor concentration, taking longer to think, blurred vision, light sensitivity, double vision and restlessness. Symptoms are rated on a 5 point scale of how much of a problem they are, from 0 (not experienced at all) to 5 (a severe problem). The maximum possible score is 64. Potter, Leigh, Wade and Fleminger (2006) suggested possible cut-offs for interpreting scores on the RPQ: scores of 0-12 as minimal, 13-24 as mild, 25-32 as moderate and 33 and above as severe.

Other questionnaires exist which are less widely used but are similar in their factor structures, for example: the Post-Concussion Syndrome Checklist (PCSC, Gouvier, Cubic, Jones, Brantley, & Cutlip, 1992), the Postconcussion Syndrome Questionnaire (PCSQ, (Axelrod, 2002; Lees-Haley, 1992) and the Post-concussions Symptoms Questionnaire (Andersson, Emanuelson, Olsson, Stålhammar, & Starmark, 2006).

2.5 The psychometric properties of the Rivermead Post-concussion Symptoms Questionnaire

King and colleagues (1995) developed the RPQ and found it to have good reliability when used as a self-report or clinician rated measure. They found that some individual items were more consistently rated than others, and that in general total score was more consistent at retest than individual items. The authors conclude "The higher reliability of the total PCS score compared with the variability of individual symptom scores may reflect constancy in the overall subjective experience of PCS by a patient regardless of variation in specific symptoms…We do not believe that the differences in reliability in some of the individual symptoms are significant enough to undermine the overall robustness of the questionnaire" (King et al., 1995, p590-591). Since this original evaluation, several others studies have investigated the psychometric

qualities of the RPQ, including reliability, validity and specificity of PCS symptoms measured by the RPQ with various conclusions and suggested changes to the original 16 item RPQ.

Two main studies have highlighted a lack of unidimensionality in the RPQ. Eyres, Carey, Gilworth, Neumann and Tennant (2005) investigated the reliability and internal and external construct validity of the RPQ using Rasch analysis with a sample of patient 3-6 months post-injury. The authors found poor internal construct validity and that three items in particular did not fit to the Rasch model: headaches, dizziness and nausea. They conclude that these items should be analysed separately from the other 13 items on the RPQ. Once the RPQ is split into these two scales ('RPQ-3' and 'RPQ-13') these have good test-retest reliability and adequate external construct validity. However, it should be noted Rasch analyses are a function of their sample and no similar studies have been conducted in a sample of people with persistent PCS symptoms beyond 6 months. Lannsjö, Borg, Björklund, Af Geijerstam, & Lundgren-Nilsson (2011) further investigated the internal construct validity of the RPQ in a homogenous sample of patients followed up 3 months after a MTBI. Similarly to Eyres et al. (2005), they concluded that the RPQ total score should not be used and that the scale is not one-dimensional and that the RPQ may not be optimal for use in the MTBI population for these reasons.

Smith-Seemiller et al. (2003) compared scores on the RPQ from a group of Chronic Pain patients to a group of patients with MTBI and found that total scores did not differ significantly. However, when symptoms were separated out into 3 factor groups of symptoms (emotional, somatic and cognitive) there was a significant difference between the groups on cognitive items, with MTBI patients significantly more likely to endorse cognitive items than chronic pain patients. They suggest using this three factor model in place of the full score.

Potter et al. (2006) used structural equation modelling to investigate the structure of PCS symptoms as measured by the RPQ. They used a one factor solution (the total RPQ score), the three factor solution suggested by Smith-Seemiller et al. (2003) and

also a two factor solution combining emotional and somatic symptoms. Their results did not support the use of the total RPQ score (as in previously discussed studies), and found the two and three factor solutions to better fit the data. Herrmann et al.'s (2009) factor analysis also supported the 3 factor model over using the total score. Another critique of the RPQ comes from a study by Laborey et al. (2014) who found only six symptoms on the RPQ to be clearly specific to MTBI (headaches, dizziness, forgetfulness, poor concentration, taking longer to think and blurred vision).

2.6 The current study

In spite of the apparent popularity of the RPQ, with citations in over 240 papers, its ability to correctly classify individuals with and without post-concussional symptoms has not been studied; previously proposed cut-offs (Potter et al., 2006) were established only on the basis of cumulative percentages of total RPQ scores. The current study aims to evaluate the specificity and sensitivity of the RPQ in identifying individuals with persistent PCS versus healthy adults. The study is warranted due to the importance of correctly classifying people with PCS and without and to enable the service to use the RPQ in the most optimal way. Given that previous studies investigating the psychometric properties of the RPQ have raised questions about its unidimensionality and suggested alternative approaches other than using the total score, this study will explore the sensitivity and specificity of the different versions of the RPQ proposed by previous research: the full RPQ, RPQ-13, RPQ-3 and 3-factor RPQ. We also aim to compare the classification accuracy of the RPQ at the existing and optimal cut-offs, and compare the different versions of the RPQ with each other.

3. Methodology

The study was approved by the South London and Maudsley NHS Foundation trust Clinical Audit & Effectiveness committee.

3.1 Participants

The participants for this study comprised two groups of participants whose self-report data had been collected by previous studies at the Lishman Unit (a longitudinal randomised controlled trial on the effectiveness of CBT for persistent PCS, and a

cross-sectional study examining aspects of symptom appraisals and the good old days bias) and who gave consent for their data to be used anonymously for future research. The first group consisted of patients with PCS as defined below (n=61) and the second group were healthy controls (n=46).

PCS participants met the following criteria: (1) individuals had sustained a mild to moderate TBI, defined using the ACRM criteria (2) symptoms consistent with the DCR ICD-10 diagnostic criteria for post-concussional disorder (see Appendix 2) (3) the head injury occurred at least six months ago (i.e. symptoms were persistent). In addition, individuals were required to be (4) current or past service users at one of the co-operating sites; (5) fluent in English (6) at least 18 years old and (7) able to give free and informed consent. Individuals were excluded from the study if they had sustained a severe TBI, defined as any TBI associated with PTA of greater than 7 days or one requiring active neurosurgical intervention (e.g. craniotomy). Individuals were also excluded if they had a prior history of neurological conditions, such as epilepsy, which may have accounted for their symptoms.

Healthy control participants met the following criteria: (1) fluent in English (2) at least 18 years of age and (3) able to give free and informed consent. Healthy control participants were excluded from the study if they had a history of any form of traumatic brain injury (that would meet at minimum the MTBI ACRM criteria described above), or a self-reported history of psychiatric disorder.

3.2 Procedure and analyses

Relevant data (demographics, RPQ data, HADS) was extracted from existing databases. Duplicate participants who had participated in both studies were excluded, along with participants for whom there was missing data from RPQ items or demographics. RPQ scores were calculated excluding items scored as 0 (never experienced) or 1 (no more of a problem).

The accuracy of the RPQ in correctly classifying PCS patients was investigated by conducting a receiver-operating characteristic (ROC) curve analysis. The accuracy of a

diagnostic test is characterized by its sensitivity and specificity. The sensitivity and specificity of a test, however, depends on the level that has been chosen as the cut-off point for normal or abnormal. A ROC analysis is widely accepted as a method for selecting an optimal cut-off point for a test and for comparing the accuracy of diagnostic tests. The overall accuracy of a diagnostic scheme is based on the area under the curve (AUC), such that an area of .50 represents classification at chance levels and an area of 1.00 represents perfect classification. Therefore the curve represents the ability of the instrument or tests to discriminate between 'cases' and 'non-cases' across the total spectrum of morbidity, and the AUC can be used to assess the discriminating ability of a screening instrument (Swets, 1986). Swets (1988) suggests heuristically interpreting AUC values as small (>0.5 to ≤0.7), moderate (>0.7 to ≤0.9), or high (>0.9 to ≤1). Precise interpretation of the accuracy of the diagnostic criteria depends on the test threshold used to determine a positive test (i.e., number of symptoms endorsed, total score). The Youden index (J) is also commonly used to measure the diagnostic effectiveness of a tool (Faraggi, 2000; Youden, 1950; Ruopp, Perkins, Whitcomb, & Schisterman, 2008). Youden's J ranges between 0 and 1, with values closer to 1 indicating that the effectiveness of the measure is relatively large. J is defined as: Specificity + sensitivity – 1. Therefore J occurs at the optimal cut-off point, maximising the number of correctly classified patients. However, depending on the clinical consequences of a diagnosis, or indeed the consequences of the exclusion patients who actually do have the disorder, this can be balanced.

Analyses were completed in SPSS v22 (IBM Corp., 2013). ROC analyses were run on the full RPQ, the RPQ 3 and 13, and the 3 factor RPQ. Comparisons between AUCs of different versions of the RPQ and Youden's J indices were computed in MedCalc v12.7.8 (MedCalc Software, Ostend, Belgium). Statistical significance was set at $p<0.05$.

4. Results

There was not a statistically significant difference between the PCS and control groups in terms of gender (49% female vs 67% female). There was a significant difference in age between the PCS group and controls, with PCS patients typically older (see *Table 1*).

Within the PCS group, 43 participants had a mild TBI, 16 had a moderate TBI and 2 could not be classified due to length of PTA unknown. The average length of post-traumatic amnesia in the PCS group was 25.4 hours (SD=39.4). The most common cause of injury was a road traffic accident (53.5%), followed by other causes including accidents and falls (34.4%) with assault the least common (13.1%).

Table 1 shows the mean RPQ scores for controls and PCS patients on the full RPQ, the RPQ-13 and the 3-factor version of the RPQ. On the full RPQ and RPQ-13 PCS patients scored significantly higher than controls. On the 3 factor version, PCS patients scored on average significantly higher on average on the cognitive, emotional and somatic factors. There was no significant difference between total RPQ scores for males and females, nor on any individual items. There was a trend for females to score higher on the headaches (p=0.06) and forgetfulness (p=0.07) items. On the HADS, PCS patients scored significantly higher than the control group, and on average scored above the suggested clinical cut-off of 8 for possible depression (Zigmond & Snaith, 1983).

Table 1: Demographic comparisons and scores on full RPQ, RPQ-13 and 3 factor RPQ for controls and PCS patients

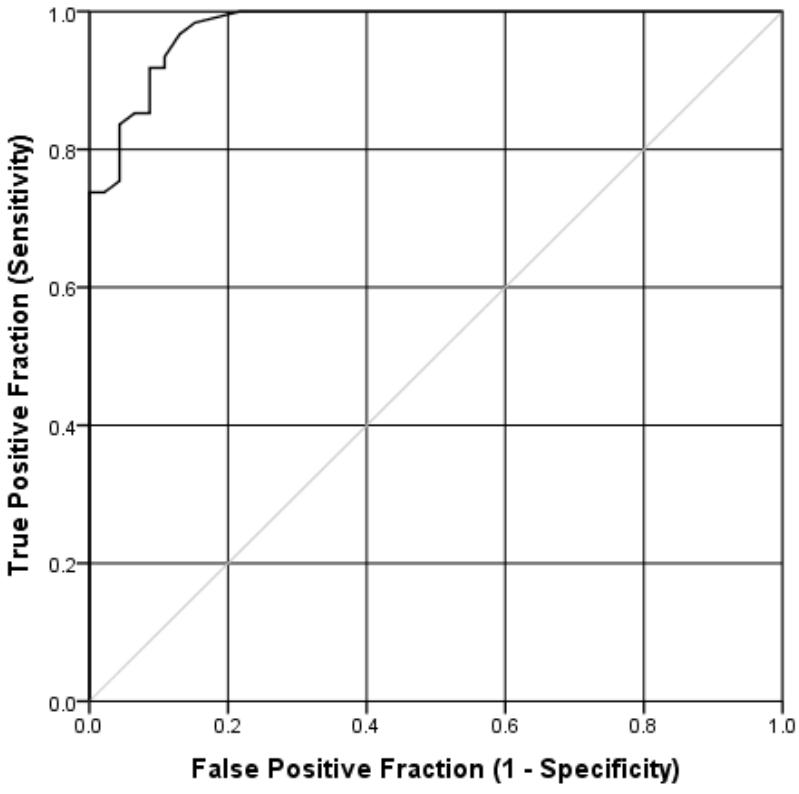
	PCS patients M (SD)	Controls M (SD)	t (df)	p
Age	41.8 (12.1)	36.3 (11.8)	-2.38 (105)	0.019
Mean Total RPQ score	34.79 (10.23)	9.59 (6.7)	15.37 (103.2)	<0.001
Mean Total RPQ 13 score	29.66 (8.74)	7.9 (6.15)	15.1 (104.5)	<0.001
Mean RPQ Cognitive items	3.16 (0.77)	0.62 (0.68)	17.79 (105)	<0.001
Mean RPQ Emotional items	2.44 (1.04)	0.72 (0.66)	10.42 (102.1)	<0.001

Mean RPQ Somatic items	1.73 (0.72)	0.58 (0.47)	9.91 (101.9)	<0.001
Mean HADS Depression total score	9.80 (3.91)	2.24 (2.61)	11.95 (103.7)	<0.001

4.1 ROC analysis of the full RPQ

The first ROC analysis assessed the ability of the full 16 item RPQ total score to correctly identify 61 patients with PCS (as determined by external criteria) from the total sample which included controls without PCS or head injury. This analysis revealed a ROC curve for the RPQ total score (AUC=0.978, p<0.0001, SE=0.011, 95% CI 0.957-0.999) that was significantly better than chance in correctly classifying individuals with RPQ versus controls (See Figure 1) and considered to be of high classification accuracy.

Figure 1: ROC Curve – correct classification by full RPQ



From the coordinates of the ROC curve, potential cut-off points were selected. *Table 2* shows the different potential cut-offs and their characteristics in terms of specificity vs. sensitivity. We have included the Youden's J index scores to aid selection of a cut-off, according to the priorities of the service. J occurs in this test at 16, indicating scores over 16 as the optimal cut-off point. However, Youden's J at 19.5 is almost identical, therefore this could also be used as a cut-off score.

Table 2: Potential cut-off points for full RPQ

Cut-off score	>12*	>13.5	>16**	>17.5	>19.5
True Positive n = (%)	61 (100%)	61 (100%)	59 (96%)	57 (93%)	56 (92%)
False Positive n = (%)	15 (23%)	10 (22%)	6 (13%)	5 (11%)	4 (9%)
True Negative n = (%)	31 (77%)	36 (78%)	40 (87%)	41 (89%)	42 (91%)
False Negative n = (%)	0	0	2 (4%)	4 (7%)	5 (8%)
Sensitivity	1	1	.967	.934	.918
1-Specificity	.239	.217	.130	.109	.087
Youdens-J	0.761	.783	.837**	.826	.831***

**Current suggested cut-off for mild symptoms (Potter et al., 2006).*

***Youden's J; optimal cut-off.*

****Possible alternative cut-off*

4.2 ROC analysis of the RPQ-13 and RPQ-3

The second ROC analysis assessed the sensitivity and specificity of the RPQ-13, with 3 items (headaches, dizziness and nausea) removed as suggested by (Eyres et al., 2005). This analysis revealed a ROC curve for the RPQ-13 (AUC=0.973, p<0.0001, SE=.012, 95% CI 0.950-0.997) that was significantly better than chance in classifying individuals with RPQ versus controls (See Figure 2) and of equivalent accuracy as the full RPQ (high classification accuracy).

A ROC analyses was also run on the 3 removed items (RPQ -3) as suggested by (Eyres et al., 2005). This analyses revealed that the ROC curve for these 3 items combined was still significantly better than chance at differentiating the groups (AUC=0.790, p<0.0001, SE=0.044, 95% CI 0.704-0.876) but with lower classification accuracy than the full RPQ and the RPQ-13. These 3 items would be considered to have moderate classification accuracy.

Figure 2: ROC Curve – correct classification by the RPQ-13 and RPQ-3

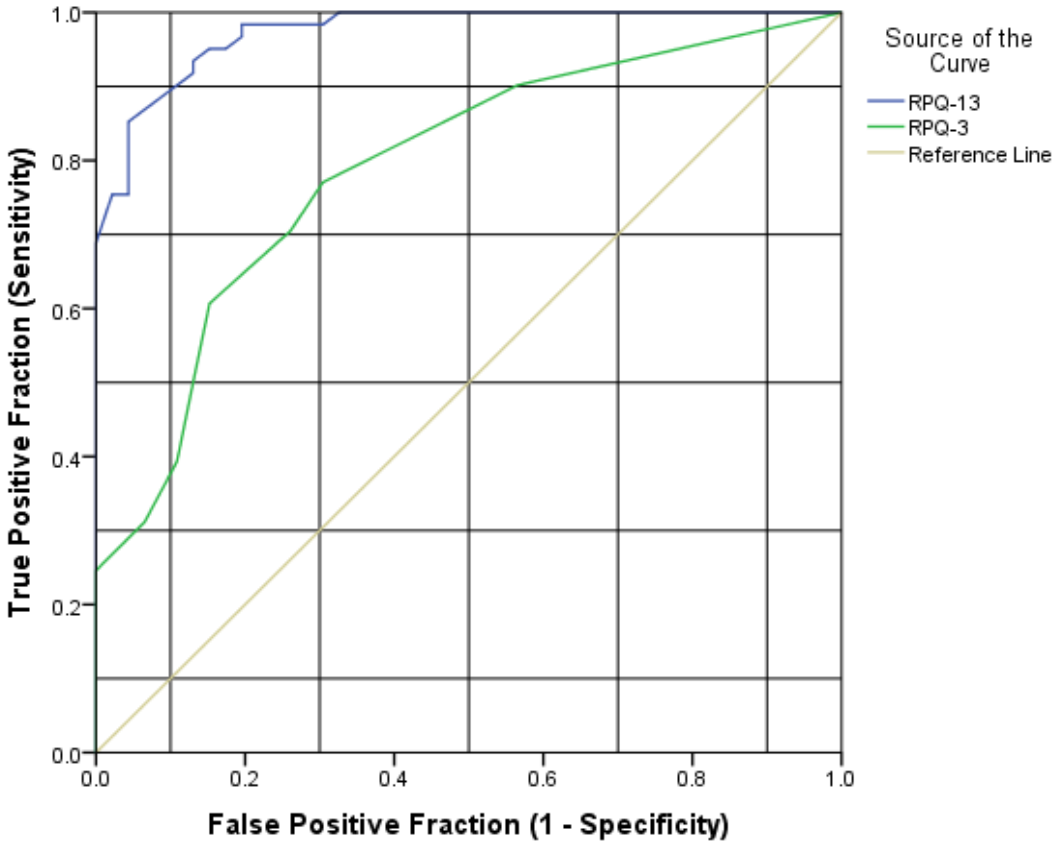


Table 3 below describes the possible cut-off points for the RPQ-13. Youden's J suggests a cut-off of 20 points, although a lower cut-off is more sensitive.

Table 3: Potential cut-off points for RPQ-13

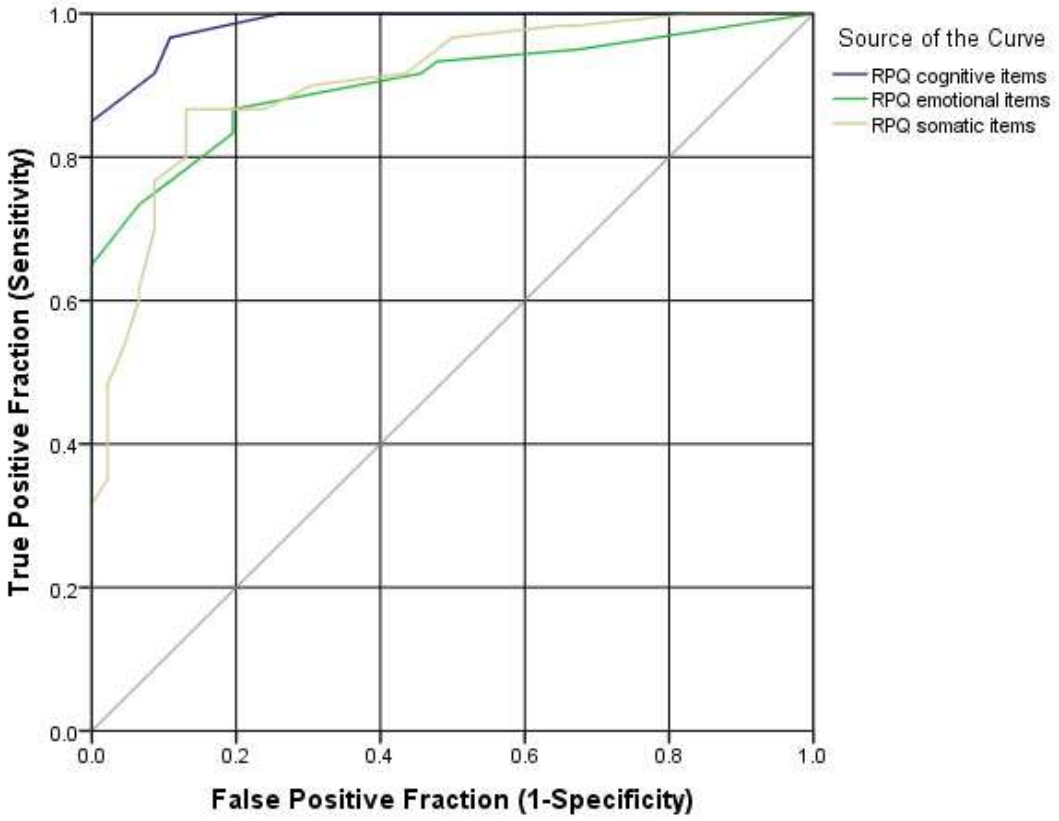
Cut-off score	<12	<15	<17	<20*
True Positive n=(%)	58 (95%)	57 (93%)	55 (90%)	52 (85%)
False Positive n=(%)	8 (17%)	6 (13%)	5 (11%)	2 (4%)
True Negative n=(%)	38 (83%)	40 (87%)	41 (89%)	44 (96%)
False Negative n=(%)	3 (5%)	4 (7%)	6 (10%)	9 (15%)
Sensitivity	.951	.934	.902	.853
1-Specificity	.174	.130	.109	.044
Youdens-J	0.777	0.804	0.793	.809*

**Youden's J; optimal cut-off.*

4.3 ROC analysis of the 3-Factor RPQ

A ROC analysis was run on the '3 factor RPQ', which separates out cognitive, emotional and somatic items, as suggested by Smith-Seemiller et al. (2003) and supported by Herrmann et al. (2009) and Potter et al. (2006). Figure 3 demonstrates that the 3 factors had differing AUCs, with the cognitive factor performing best (AUC=0.986, p<0.001, SE=0.008, 95%CI 0.971-1) with high classification accuracy. For the somatic factor the AUC was 0.902 (p<0.001, SE=0.030, 95%CI 0.849-0.962) and for the emotional factor AUC was 0.905 (p<0.001, SE=0.029, 0.843-0.961), both with moderate classification accuracy.

Figure 3: ROC curve – Correct classification by RPQ 3-factor



4.4 Comparisons between AUCs

Table 4 displays the comparisons between the ROC curves. The AUCs of the full RPQ and the RPQ-13 were not significantly different from each other (p=0.48, SE=0.007). The difference in AUCs between the full RPQ and the RPQ-13 versus the somatic and emotional factors were significantly different (p=<0.05), with the full RPQ and RPQ-13 significantly out-performing either factor alone. There was not a significant difference between the full RPQ and the RPQ-13 versus cognitive items alone (p=0.35 and p=0.14 respectively).

Table 4: Pairwise comparison of ROC curves

	Difference between AUC	Significance level
RPQ full vs RPQ 13	0.0049	P = 0.485
RPQ full vs RPQ Cognitive	0.0074	P = 0.353
RPQ full vs RPQ Emotional	0.0764	P = 0.003*
RPQ full vs RPQ Somatic	0.0734	P = 0.002*
RPQ-13 vs RPQ Cognitive	0.0123	P = 0.143
RPQ-13 vs RPQ Emotional	0.0716	P = 0.002*
RPQ-13 vs RPQ Somatic	0.0685	P = 0.003*
RPQ Cognitive vs RPQ Emotional	0.0839	P = 0.003*
RPQ Cognitive vs RPQ Somatic	0.0808	P = 0.002*
RPQ Emotional vs RPQ Somatic	0.0031	P = 0.935

**significant*

5. Discussion

5.1 Discussion of results

There is much debate about the validity of a post-concussion syndrome 'diagnosis'. Nonetheless, many people can, and do, suffer symptoms which are attributed to the after effects of a MTBI, and can be very disabling. A World Health Organisation (WHO) task force (Carroll, Cassidy, Holm, Kraus, & Coronado, 2004) reviewed the complex and conflicting literature on the nature of persistent symptoms after MTBI and identified accurate diagnosis of PCS as a priority. They concluded that predictors of long-term outcomes of MTBI also require further study, a call which researchers have

answered in recent years (e.g. Ganti et al., 2014; Sigurdardottir, Andelic, Roe, Jerstad, & Schanke, 2009). Furthermore, preliminary data from a trial of CBT for PCS by Potter et al. (submitted) suggest that PCS symptoms are amenable to psychological treatment and that CBT can reduce PCS symptoms, as measured by the RPQ, and improve quality of life. Therefore it is important for clinicians working in the field to have an adequate tool for screening for cases of PCS, but one which does not over-estimate the likelihood of PCS when their symptoms may either be in the normal range, or which are more appropriately explained by other problems such as depression. The current study aimed to evaluate the sensitivity and specificity of the RPQ, a self-report measure of PCS, and identify an appropriate cut-off for identifying PCS in patients at the Lishman Brain Injury Unity, and for wider dissemination to other TBI services.

The results of this study suggest that the RPQ is an accurate tool for identifying PCS, with AUC in the ROC analysis of over .9 (Swets and Pickett (1982) recommended that measures with an AUC of .7-.9 can be used for diagnosis). The sensitivity and specificity of the full RPQ was good, with an optimal cut-off of 16 capturing most cases of PCS (in this study, 96% were correctly identified). Importantly, the analysis showed that the current cut-off of 12 is most likely too low, and would 'over-diagnose' PCS, with a false positive rate of 32%. 19.5 could be used as an alternative higher cut-off, as the Youden's index was almost identical. We chose to emphasise specificity to avoid false positives as much as possible, given the literature on non-specific symptoms of PCS and research which shows that patients without a TBI can score highly on the questionnaire. As can be observed, even with the higher cut-off, false positive rate was still 13% in our sample. Higher and lower cut-off data has been included to allow professionals to select one appropriate to their service or for research purposes. For the RPQ-13 a cut-off of 20 was identified. The RPQ-13 has 3 of the most sensitive items removed, and thus requires a higher cut-off. Prior to this study there was no established cut-off point for the RPQ-13, so this will be of value to clinicians wishing to use this more specific version of the RPQ as suggested by Eyres et al. (2005).

The identification of the cut-off on the RPQ is important, given the non-specificity of many PCS symptoms and the role that appraisals of symptoms can play in maintaining PCS. The consequences of wrongly diagnosing someone with PCS might include iatrogenic effects such as more severe symptoms, offering inappropriate and ineffective treatment (such as treatment for PCS when treatment for depression would be more appropriate), and could affect medico legal proceedings. The RPQ may be already prone to 'over-diagnose' PCS, as many of the symptoms it measures are experienced commonly in the general population. Indeed, in our sample most controls without TBI endorsed at least a two items on the RPQ, with an average score of 9.6. However, the analysis demonstrated that the RPQ has high classification accuracy in separating controls from people with PCS. What is less known is whether the PCS groups symptoms were better explained by other problems, for example depression or medico legal involvement, which we did not control and which may affect the expression of PCS symptoms (Carroll et al., 2004). Our sample had high levels of probable depression indicated by HADS scores, which further highlights the importance of an accurate measure to identify PCS, as it may well be mistaken for depression and vice versa. At the Lishman Brain Injury unit, both the RPQ and the HADS are routinely administered to patients who present to the service for assessment and treatment of the after effects of TBI and scores on both are considered along with interview assessment in treatment planning.

This study also examined the different versions of the RPQ suggested by previous studies. It was found that the full RPQ and the RPQ-13 were of almost equivalent sensitivity and specificity (both with high classification accuracy) indicating that the removal of the 3 items did not substantially affect the identification of cases with PCS. However, the 3 removed items when analysed separately did not perform as well, and thus, as suggested by Eyres et al. (2005), may be more prone to mis-classification. It is possible these removed items reflect symptoms more prominent early after injury (headaches, dizziness and nausea being common after head injury) or that a shorter subscale may be less sensitive and/or specific than a total score. However, what was particularly striking was that from the 3-factor model, the 3 cognitive items on the RPQ alone had almost equivalent discriminatory power to the RPQ full and the RPQ-

13, and there was no significant difference in AUC between the full RPQ, the RPQ-13 and the cognitive factor. Thus, 3 cognitive items alone could accurately distinguish between cases of PCS and controls. This finding fits with previous studies showing that PCS patients are more likely to report cognitive difficulties than other patient and control groups (e.g. Gordon, Haddad, Brown, Hibbard, & Sliwinski, 2000; Smith-Seemiller et al., 2003) and the treatment model of PCS as being characterised by subjective cognitive difficulties (Potter & Brown, 2012). The somatic and emotional symptoms were less accurate at distinguishing between cases and controls but still performed significantly better than chance.

The current participant sample differed from previous studies which often controlled for length of time since injury (e.g. Laborey et al. 2014, 3 months after injury). Our group was more heterogeneous and also had a higher mean score (34) on the RPQ than might be expected in an average TBI group, as they were selected on the basis of meeting external criteria for PCS.

5.2 Limitations

The current study has several limitations. Firstly, the sample size was relatively small compared to previous studies investigating the psychometric properties of the RPQ. Bachmann, Puhan, ter Riet and Bossuyt (2006) suggest that sample sizes in sensitivity and specificity analysis studies are often too small, and recommend that confidence intervals should be reported, as we have done. Secondly, the current study compared PCS patients to controls with no psychiatric history or experience of TBI. In reality a clinician would more likely be working within a TBI population with or without PCS. The TBI population is often complex with comorbid difficulties which may impact on their presentation. The picture would be further complicated by the lack of specificity of PCS symptoms (e.g. overlap with depression), and this study is limited by looking at these two more clearly separable groups. We did not look at disentangling patients with depression or other clinical problems versus PCS and would not generalise the results to these groups.

A related limitation with the samples used was that our PCS group came from a national tertiary service, and hence may not be representative of the typical TBI population seen in primary or secondary care services. Our patients would have been likely to have had long standing and complex complaints after a TBI. Additionally we did not control for length of time since injury or medico legal involvement which have been proposed to be factors which may influence the severity and persistence of PCS symptoms (e.g. Bazarian et al., 1999; Binder & Rohling, 1996; Fee & Rutherford, 1988).

Finally, the ROC analysis identified an optimal cut-off for separation of PCS from non-PCS scores, but did not categorise severity of PCS i.e. mild, moderate and severe. This could be a useful distinction for clinicians or researchers working with the syndrome, for example in investigating which levels of severity respond best to treatment and whether severity is associated with functional disability.

5.3 Implications for service and wider clinical practise

The current study has various implications for the Lishman service and wider clinical practise in the area. The findings suggest the Lishman Unit should modify the use of the RPQ in several ways. The RPQ is routinely administered at assessment, and previously a score of above 12 would be flagged up in the assessment report as suggesting the presence of PCS. This should be changed above 16 for the full RPQ and above 20 for the RPQ-13. In turn, for those referred for treatment of problems related to TBI, this higher cut-off should be considered in formulation of their difficulties i.e. whether they are likely part of PCS or better formulated as reflecting problems such as depression or chronic pain. If a patient has a high score on the RPQ then a course of treatment for PCS should be considered, if this is in line with the patients' priorities for treatment. An important part of such treatment will include psycho-education and intervention related to subjective cognitive difficulties. The service could also monitor outcomes of treatment on the RPQ, and which treatments help to reduce symptoms of PCS. Another potential impact which this study may have on the service is that it suggests that shorter versions of the RPQ (RPQ-13, or cognitive items only) can

distinguish PCS cases as well as the full version, and thus these could be considered given time restraints in already lengthy neurocognitive assessments.

For wider clinical practise in other TBI services, the above suggestions could also be considered, particularly the higher cut-off. We have selected a score of 16 to balance specificity and sensitivity, but have included alternative higher and lower thresholds in this report which could also be used. As has been discussed, PCS is a controversial construct and the diagnostic criteria are lacking in specificity (Boake et al., 2005, Kashluba et al., 2006) and thus there may be an argument that cut-offs could be selected according to service demands. For example, in a wider screen of the presence of significant PCS symptoms, a cut-off with higher sensitivity could be selected in order not to miss cases. Conversely, in a service with limited resources to offer treatment, specificity could be prioritised to ensure that resources are allocated only to patients with the highest need of a specialist service or in research with strict inclusion criteria.

In any service, it is important to hold in mind that PCS is a construct and accurate diagnosis is difficult to make. Even the full ICD-10 diagnostic criteria has been highlighted as lacking specificity (Kashluba et al., 2006). There are many factors which can play a role in contributing towards PCS symptoms, such as depression which may elevate scores, and individual formulation is key in providing the appropriate treatment, for example treatment of depression versus treatment of PCS.

5.4 Future recommendations

A useful future study would be to replicate the current study with a larger sample of people with enduring PCS, possibly from primary or secondary care services where the population may be more representative of typical PCS cases. This could be extended to comparing the accuracy of the RPQ to distinguish different samples of patients, for example mild TBI with PCS vs mild TBI without PCS. In the current study we compared people with a TBI to controls who had never experienced a TBI, but as has been discussed the challenge for clinicians is often distinguishing between clinical groups, for example differentiating whether someone is depressed or has PCS. This would be a

particular area of interest for future study, given the relatively high HADS scores in our PCS sample. A ROC analysis could be run comparing a depressed group and a PCS group for the classification accuracy of the RPQ.

6. Dissemination

The results of the study were presented to the South Thames Neuropsychology special interest group which is a regular meeting for psychologists working in London, Surrey and Kent with an interest in neuropsychology. Approximately 15 clinicians were present from a range of services including brain injury and neuro rehabilitation services. The presentation spread awareness of the availability of the RPQ and different ways in which it can be utilised. Clinicians were interested in the RPQ and, despite its wide use in a research context, some of them were not previously familiar with the measure. Clinicians indicated that they currently usually used clinical interviews as a screen for PCS and this the use of the RPQ as an alternative could saves clinicians valuable assessment time in the future. Clinicians commented on the difficulty of distinguishing post-concussion symptoms from other disorders such as depression, which we have highlighted as an area for future study, and the importance of having accurate screening procedures.

One query was raised regarding whether there is an informant (e.g. carer) rated version of the RPQ. This would be an interesting area for future study, as it may be that informant and self-report vary. Informants may report less severe symptoms, given that PCS is often characterised by hyper awareness of symptoms and interpretation by the patient. An example of this is the 'good old days bias' (Gunstad & Suhr, 2004; Iverson, Lange, Brooks, & Rennison, 2010; Lange, Iverson, & Rose, 2010), where people who have suffered MTBI underestimate the presence of past problems and therefore may overestimate the severity of current problems.

The results of the study were also fed back directly to the psychology team at the Lishman Brain Injury Unit and will influence subsequent scoring of the RPQ and identification of the possible PCS symptoms in outpatients with MTBI, which in turn will affect the formulation and treatment options offered to these patients, for example whether they are offered CBT for PCS which is in development in the service.

The project also is currently being written up, with the assistant psychologist from the team, to be submitted to a peer reviewed academic journal for publication so that results are disseminated more widely among health professionals and researchers working with patients with PCS symptoms. Thus this study has the potential to impact on the diagnosis, formulation and treatment of PCS in the TBI population where the RPQ is used as the measure of choice. A poster presentation of this study was presented at the Acquired Brain Injury Forum for London (ABIL) on 9th December 2014. A report of the study will also be available in the archives of the South London and Maudsley NHS Foundation trust Audit department.

7. Conclusion

In conclusion, the present study evaluated the sensitivity and specificity of the RPQ, a questionnaire used by the Lishman Brain Injury service as a screen for PCS, but for which there was no previously identified cut-off score. It is important for the service to correctly identify cases of PCS for assessments which may be used in legal proceedings, and to inform formulations and treatment plans within the service, i.e. whether treatment will involve exploration of post-concussion symptoms. This study found the RPQ to be a good tool for distinguishing PCS patients from controls, and that for the full RPQ a cut-off of 16 could be used. The shorter RPQ-13 item and the cognitive items of the 3 factor model of the RPQ also had good classification accuracy. Further research recommendations would be to conduct a similar analysis comparing known PCS patients and other clinical groups and to study patients within a primary or secondary tier service, which may be more representative of 'typical' TBI patients. Results were disseminated to other clinicians working with TBI in south London and will be submitted for publication in a peer reviewed journal.

8. References

American Congress of Rehabilitational Medicine. (1993). Definition of mild traumatic brain injury. *Journal of Head Trauma Rehabilitation*, *8*, 89–87.

Anderson, T., Heitger, M., & Macleod, A. D. (2006). Concussion and mild head injury. *Practical Neurology*, *6*(6), 342–357. doi:10.1136/jnnp.2006.106583

Andersson, E., Emanuelson, I., Olsson, M., Stålhammar, D., & Starmark, J.-E. (2006). the New Swedish Post-Concussion Symptoms Questionnaire: a Measure of Symptoms After Mild Traumatic Brain Injury and Its Concurrent Validity and Inter-Rater Reliability. *Journal of Rehabilitation Medicine*, *38*(1), 26–31. doi:10.1080/16501970500310564

Axelrod, B. (2002). Construct validity of the PCSQ as related to the MMPI-2. *Archives of Clinical Neuropsychology*, *17*(4), 343–350. doi:10.1016/S0887-6177(01)00119-6

Bachmann, L. M., Puhan, M. A., ter Riet, G., & Bossuyt, P. M. (2006). Sample sizes of studies on diagnostic accuracy: literature survey. *BMJ (Clinical Research Ed.)*, *332*(7550), 1127–9. doi:10.1136/bmj.38793.637789.2F

Bazarian, J. J., Wong, T., Harris, M., Leahey, N., Mookerjee, S., & Dombovy, M. (1999). Epidemiology and predictors of post-concussive syndrome after minor head injury in an emergency population. *Brain Injury*, *13*(3), 173–189. Retrieved from http://informahealthcare.com/doi/abs/10.1080/026990599121692

Binder, L. M., & Rohling, M. L. (1996). Money Matters: A Meta-Analytic Review of the Effects of Financial Incentives on Recovery After Closed-Head Injury. *American Journal of Psychiatry*, *153*(1), 7–10.

Boake, C., McCauley, S. R., Levin, H. S., Pedroza, C., Contant, C. F., Song, J. X., … Diaz-Marchan, P. J. (2005). Diagnostic criteria for postconcussional syndrome after mild to moderate traumatic brain injury. *The Journal of Neuropsychiatry and Clinical Neurosciences*, *17*(3), 350–6. doi:10.1176/appi.neuropsych.17.3.350

Carroll, L., Cassidy, J. D., Holm, L., Kraus, J., & Coronado, V. (2004). Methodological issues and research recommendations for mild traumatic brain injury: the who collaborating centre task force on mild traumatic brain injury. *Journal of Rehabilitation Medicine*, *36*(2), 113–125. doi:10.1080/16501960410023877

Chan, R. C. K. (2001). Base rate of post-concussion symptoms among normal people and its neuropsychological correlates. *Clinical Rehabilitation*, *15*(3), 266–273. doi:10.1191/026921501675253420

Eyres, S., Carey, A., Gilworth, G., Neumann, V., & Tennant, A. (2005). Construct validity and reliability of the Rivermead Post-Concussion Symptoms Questionnaire. *Clinical Rehabilitation*, *19*(8), 878–887. doi:10.1191/0269215505cr905oa

Faraggi, D. (2000). The effect of random measurement error on receiver operating characteristic (ROC) curves. *Statistics in Medicine*, *19*(1), 61–70. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/10623913

Fee, C. R., & Rutherford, W. H. (1988). A study of the effect of legal settlement on post-concussion symptoms. *Archives of Emergency Medicine*, *5*(1), 12–7. Retrieved from http://www.pubmedcentral.nih.gov/articlerender.fcgi?artid=1285470&tool=pmcentrez&rendertype=abstract

Ganti, L., Khalid, H., Patel, P. S., Daneshvar, Y., Bodhit, A. N., & Peters, K. R. (2014). Who gets post-concussion syndrome? An emergency department-based prospective analysis. *International Journal of Emergency Medicine*, *7*(31), 1–7.

Gordon, W. A., Haddad, L., Brown, M., Hibbard, M. R., & Sliwinski, M. (2000). The sensitivity and specificity of self-reported symptoms in individuals with traumatic brain injury. *Brain Injury : [BI]*, *14*(1), 21–33. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/10670659

Gouvier, W. D., Cubic, B., Jones, G., Brantley, P., & Cutlip, Q. (1992). Postconcussion symptoms and daily stress in normal and head-injured college populations. *Archives of Clinical Neuropsychology : The Official Journal of the National Academy of Neuropsychologists*, *7*(3), 193–211. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/14591254

Gunstad, J., & Suhr, J. A. (2004). Cognitive factors in Postconcussion Syndrome symptom report. *Archives of Clinical Neuropsychology : The Official Journal of the National Academy of Neuropsychologists*, *19*(3), 391–405. doi:10.1016/S0887-6177(03)00073-8

Herrmann, N., Rapoport, M., Rajaram, R., Chan, F., Kiss, A., Ma, A., … Lanctôt, K. (2009). Factor Analysis of the Rivermead Post-Concussion Symptoms Questionnaire in Mild-to-Moderate Traumatic Brain Injury Patients. *The Journal of Neuropsychiatry and Clinical Neurosciences*, *21*(2), 181–188. Retrieved from http://journals.psychiatryonline.org/article.aspx?articleid=103723

IBM Corp. (2013). IBM SPSS Statistics for Windows, Version 22.0. Armonk, NY: IBM Corp.

Iverson, G. L. (2006). Misdiagnosis of the persistent postconcussion syndrome in patients with depression. *Archives of Clinical Neuropsychology : The Official Journal of the National Academy of Neuropsychologists*, *21*(4), 303–10. doi:10.1016/j.acn.2005.12.008

Iverson, G. L., & Lange, R. T. (2003). Examination of “postconcussion-like” symptoms in a healthy sample. *Applied Neuropsychology*, *10*(3), 137–44. doi:10.1207/S15324826AN1003_02

Iverson, G. L., Lange, R. T., Brooks, B. L., & Rennison, V. L. A. (2010). “Good old days” bias following mild traumatic brain injury. *The Clinical Neuropsychologist*, *24*(1), 17–37. doi:10.1080/13854040903190797

Kashluba, S., Casey, J. E., & Paniak, C. (2006). Evaluating the utility of ICD-10 diagnostic criteria for postconcussion syndrome following mild traumatic brain injury. *Journal of the International Neuropsychological Society : JINS*, *12*(1), 111–8. doi:10.1017/S1355617706060036

Kay, A., & Teasdale, G. (2001). Head injury in the United Kingdom. *World Journal of Surgery*, *25*(9), 1210–20. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/11571960

Kay, T. (1993). Neuropsychological treatment of mild traumatic brain injury. *Journal of Head Trauma Rehabilitation*, *September*, 74–85.

King, N. S. (2003). Post-concussion syndrome: clarity amid the controversy? *The British Journal of Psychiatry : The Journal of Mental Science*, *183*, 276–8. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/14519601

King, N. S., Crawford, S., Wenden, F. J., Moss, N. E., & Wade, D. T. (1995). The Rivermead Post Concussion Symptoms Questionnaire: a measure of symptoms commonly experienced after head injury and its reliability. *Journal of Neurology*, *242*(9), 587–92. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/8551320

Laborey, M., Masson, F., Ribéreau-Gayon, R., Zongo, D., Salmi, L. R., & Lagarde, E. (2014). Specificity of postconcussion symptoms at 3 months after mild traumatic brain injury: results from a comparative cohort study. *The Journal of Head Trauma Rehabilitation*, *29*(1), E28–36. doi:10.1097/HTR.0b013e318280f896

Lange, R. T., Iverson, G. L., & Rose, A. (2010). Post-concussion symptom reporting and the “good-old-days” bias following mild traumatic brain injury. *Archives of Clinical Neuropsychology : The Official Journal of the National Academy of Neuropsychologists*, *25*(5), 442–50. doi:10.1093/arclin/acq031

Lannsjö, M., Borg, J., Björklund, G., Af Geijerstam, J.-L., & Lundgren-Nilsson, A. (2011). Internal construct validity of the Rivermead Post-Concussion Symptoms Questionnaire. *Journal of Rehabilitation Medicine : Official Journal of the UEMS European Board of Physical and Rehabilitation Medicine*, *43*(11), 997–1002. doi:10.2340/16501977-0875

Lees-Haley, P. R. (1992). Neuropsychological complaint base rates of personal injury claimants. *Forensic Reports*, *5*, 395–391.

Lishman, W. A. (1988). Physiogenesis and psychogenesis in the “post-concussional syndrome”. *The British Journal of Psychiatry : The Journal of Mental Science*, *153*, 460–9. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/3074853

Mittenberg, W., & Burton, D. B. (1994). A survey of treatments for post-concussion syndrome. *Brain Injury : [BI]*, *8*(5), 429–37. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/7951205

Mittenberg, W., Canyock, E. M., Condit, D., & Patton, C. (2001). Treatment of post-concussion syndrome following mild head injury. *Journal of Clinical and Experimental Neuropsychology*, *23*(6), 829–36. doi:10.1076/jcen.23.6.829.1022

Mittenberg, W., Zielinski, R., & Fichera, S. (1993). Recovery from Mild Head Injury: *Psychotherapy in Private Practice*, *12*(2), 37–52. doi:10.1300/J294v12n02_03

Potter, S., & Brown, R. G. (2012). Cognitive behavioural therapy and persistent post-concussional symptoms: integrating conceptual issues and practical aspects in treatment. *Neuropsychological Rehabilitation*, *22*(1), 1–25. doi:10.1080/09602011.2011.630883

Potter, S., Brown, R. G., & Fleminger, S. (submitted). A randomised, waiting list controlled trial of cognitive-behavioural therapy for persistent postconcussional symptoms. *Journal of Head Trauma Rehabilitation*.

Potter, S., Leigh, E., Wade, D., & Fleminger, S. (2006). The Rivermead Post Concussion Symptoms Questionnaire: a confirmatory factor analysis. *Journal of Neurology*, *253*(12), 1603–14. doi:10.1007/s00415-006-0275-z

Rohling, M. L., Binder, L. M., Demakis, G. J., Larrabee, G. J., Ploetz, D. M., & Langhinrichsen-Rohling, J. (2011). A meta-analysis of neuropsychological outcome after mild traumatic brain injury: re-analyses and reconsiderations of Binder et al. (1997), Frencham et al. (2005), and Pertab et al. (2009). *The Clinical Neuropsychologist*, *25*(4), 608–23. doi:10.1080/13854046.2011.565076

Ruff, R. M., Camenzuli, L., & Mueller, J. (1996). Miserable minority: emotional risk factors that influence the outcome of a mild traumatic brain injury. *Brain Injury : [BI]*, *10*(8), 551–65. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/8836512

Ruopp, M. D., Perkins, N. J., Whitcomb, B. W., & Schisterman, E. F. (2008). Youden Index and optimal cut-point estimated from observations affected by a lower limit of detection. *Biometrical Journal. Biometrische Zeitschrift*, *50*(3), 419–30. doi:10.1002/bimj.200710415

Sayegh, A. Al, Sandford, D., & Carson, A. J. (2010). Psychological approaches to treatment of postconcussion syndrome : a systematic review, 1128–1135. doi:10.1136/jnnp.2008.170092

Sigurdardottir, S., Andelic, N., Roe, C., Jerstad, T., & Schanke, A.-K. (2009). Post-concussion symptoms after traumatic brain injury at 3 and 12 months post-injury: a prospective study. *Brain Injury : [BI]*, *23*(6), 489–97. doi:10.1080/02699050902926309

Smith-Seemiller, L., Fow, N. R., Kant, R., & Franzen, M. D. (2003). Presence of post-concussion syndrome symptoms in patients with chronic pain vs mild traumatic brain injury. *Brain Injury*, *17*(3), 199–206. doi:10.1080/0269905021000030823

Styrke, J., Sojka, P., Björnstig, U., Bylund, P.-O., & Stålnacke, B.-M. (2013). Sex-differences in symptoms, disability, and life satisfaction three years after mild traumatic brain injury: a population-based cohort study. *Journal of Rehabilitation Medicine : Official Journal of the UEMS European Board of Physical and Rehabilitation Medicine*, *45*(8), 749–57. doi:10.2340/16501977-1215

Swets, J. A. (1986). Form of empirical ROCs in discrimination and diagnostic tasks: implications for theory and measurement of performance. *Psychological Bulletin*, *99*(2), 181–98. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/3515382

Swets, J. A. (1988). Measuring the accuracy of diagnostic systems. *Science (New York, N.Y.)*, *240*(4857), 1285–93. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/3287615

Swets, J. A., & Pickett, R. M. (1982). *Evaluation of Diagnostic Systems*. Academic Press.

Tiersky, L. A., Anselmi, V., Johnston, M. V, Kurtyka, J., Roosen, E., Schwartz, T., & Deluca, J. (2005). A trial of neuropsychologic rehabilitation in mild-spectrum traumatic brain injury. *Archives of Physical Medicine and Rehabilitation*, *86*(8), 1565–74. doi:10.1016/j.apmr.2005.03.013

Wang, Y., Chan, R. C. K., & Deng, Y. (2006). Examination of postconcussion-like symptoms in healthy university students: relationships to subjective and objective neuropsychological function performance. *Archives of Clinical Neuropsychology : The Official Journal of the National Academy of Neuropsychologists*, *21*(4), 339–47. doi:10.1016/j.acn.2006.03.006

Wood, R. L. (2004). Understanding the “miserable minority”: a diasthesis-stress paradigm for post-concussional syndrome. *Brain Injury : [BI]*, *18*(11), 1135–53. doi:10.1080/02699050410001675906

Wood, R. L. (2007). Post concussional syndrome: all in the minds eye! *Journal of Neurology, Neurosurgery, and Psychiatry*, *78*(6), 552. doi:10.1136/jnnp.2006.113845

Youden, W. J. (1950). Index for rating diagnostic tests. *Cancer*, *3*(1), 32–35. doi:10.1002/1097-0142(1950)3:1<32::AID-CNCR2820030106>3.0.CO;2-3

Zigmond, A. S., & Snaith, R. P. (1983). The Hospital Anxiety and Depression Scale. *Acta Psychiatrica Scandinavica*, *67*(6), 361–70. doi:10.1111/j.1600-0447.1983.tb09716.x

9. Appendices

Appendix 1: DSM-IV criteria for Post-Concussion Syndrome:

A) History of TBI causing "significant cerebral concussion;"

B) Cognitive deficit in attention and/or memory;

C) Presence of at least three of eight symptoms (e.g., fatigue, sleep disturbance, headache, dizziness, irritability, affective disturbance, personality change, apathy) that appear after injury and persist for 3 months;

D) Symptoms that begin or worsen after injury;

E) Interference with social role functioning; and

F) Exclusion of dementia due to head trauma and other disorders that better account for the symptoms.

Appendix 2: ICD-10-DCR criteria for FO7.2 Postconcussional syndrome, WHO (1993)

A. The general criteria of F07 must be met.

B. History of head trauma with loss of consciousness, preceding the onset of symptoms by a period of up to four weeks (objective EEG, brain imaging, or oculonystagmographic evidence for brain damage may be lacking).

C. At least three of the following:

(1) Complaints of unpleasant sensations and pains, such as headache, dizziness (usually lacking the features of true vertigo), general malaise and excessive fatigue or noise intolerance.

(2) Emotional changes, such as irritability, emotional lability, both easily provoked or exacerbated by emotional excitement or stress, or some degree of depression and/or anxiety.

(3) Subjective complaints of difficulty in concentration and in performing mental tasks, and of memory complaints, without clear objective evidence (e.g. psychological tests) of marked impairment.

(4) Insomnia.

(5) Reduced tolerance to alcohol.

(6) Preoccupation with the above symptoms and fear of permanent brain damage, to the extent of hypochondriacal over-valued ideas and adoption of a sick role.

Appendix 3: Rivermead Post Concussion Symptoms Questionnaire (King et al., 1995).

The Rivermead Post-Concussion Symptoms Questionnaire

After a head injury or accident some people experience symptoms which can cause worry or nuisance. We would like to know if you now suffer from any of the symptoms given below. As many of these symptoms occur normally, we would like you to compare yourself now with before the accident. For each one, please circle the number closest to your answer.

0= Not experienced at all
1= No more of a problem
2= A mild problem
3= A moderate problem
4= A severe problem

Compared with before the accident, do you now (i.e., over the last 24 hours) suffer from:

Headaches	0	1	2	3	4
Feelings of dizziness	0	1	2	3	4
Nausea and/or vomiting	0	1	2	3	4
Noise Sensitivity, easily upset by loud noise	0	1	2	3	4
Sleep Disturbance	0	1	2	3	4
Fatigue, tiring more easily	0	1	2	3	4
Being Irritable, easily angered	0	1	2	3	4
Feeling Depressed or tearful	0	1	2	3	4
Feeling Frustrated or impatient	0	1	2	3	4
Forgetfulness, poor memory	0	1	2	3	4
Poor concentration	0	1	2	3	4
Taking longer to think	0	1	2	3	4
Blurred vision	0	1	2	3	4
Light sensitivity, easily upset by bright light	0	1	2	3	4
Double vision	0	1	2	3	4
Restlessness	0	1	2	3	4

Are you experiencing any other difficulties?

1. _____ 0 1 2 3 4
2. _____ 0 1 2 3 4